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Secondary aortoduodenal fistula following endovascular repair of inflammatory abdominal aortic aneurysm due to *Streptococcus anginosus* infection: A case report and literature review☆

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ABSTRACT

INTRODUCTION: Aortoenteric fistula is a rare but very serious complication of both surgical and endovascular abdominal aortic reconstruction. Since the advent of endovascular abdominal aortic aneurysm repair (EVAR), 20 cases of aortoduodenal fistula associated with aortic stent grafts have been reported.[1] However, only a handful has been reported following inflammatory abdominal aortic aneurysm repair. It most commonly presents with bleeding, usually from the upper gastro-intestinal tract. With recent advances in the screening, diagnosis and management of abdominal aortic aneurysms either surgically or through an endovascular approach, the diagnosis of an aortoduodenal fistula in patients with gastro-intestinal bleeding must be suspected and excluded.
PRESENTATION OF CASE: We describe a case of secondary aortoduodenal fistula that occurred two and a half years following endovascular stent graft repair of an inflammatory abdominal aortic aneurysm. We also outline the emergency correction plan and the attempts at repair.
DISCUSSION: This case defies the general concept that patients with inflammatory abdominal aortic aneurysms are relatively immune to rupture. Although the presence of a peri-aneurysm thick inflammatory membrane decreases the possibility of rupture, these patients are more susceptible to other related complications such as aorto-enteric and aorto-caval fistulas.[2] This case also demonstrates the peculiar presence of *Streptococcus anginosus* as the pathological organism leading to graft infection and subsequent fistula, as opposed to enterococci which are often found in endograft infection.
CONCLUSION: Aorto-enteric fistulas are associated with a grave prognosis. Early diagnosis is crucial and extra vigilance should be taken in cases of inflammatory AAA.
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1. Case Report

A 75 year-old male patient, who underwent endovascular repair of a 5.8 cm inflammatory abdominal aortic aneurysm two and a half years previously (Fig. 1), was transferred from another centre complaining of acute abdominal pain and haematemesis of one day duration. He had several co-morbidities including chronic obstructive airways disease, hypertension and a cerebrovascular accident. Upon arrival, he was shocked, confused, pale and tachycardic. His blood pressure was 90/40, heart rate 115 bpm and temperature 36 °C. On examination, his abdomen was rigid with inaudible bowel sounds, and there was no melena by digital rectal examination. His haemoglobin was 8.4 g/dL.

An oesophago-gastro-duodenoscopy (OGD) performed before transfer showed blood in the stomach down to the second part of the duodenum, the source of which could not be identified. A computerized tomography angiogram of the aorta showed multiple bubbles of gas within the aneurysm sac surrounding the stent graft (Fig. 2) with no discernible wall separating the aneurysm sac from the lumen of the distal third part of the duodenum, along with a psoas abscess measuring 6 × 4 cm.

As soon as the patient arrived, he was resuscitated, received 3 units of packed RBC's and was sent to the operating theatre for OGD. Initially, the OGD found blood clots in the stomach which obscured the field, so a laparotomy was conducted in an attempt to localize

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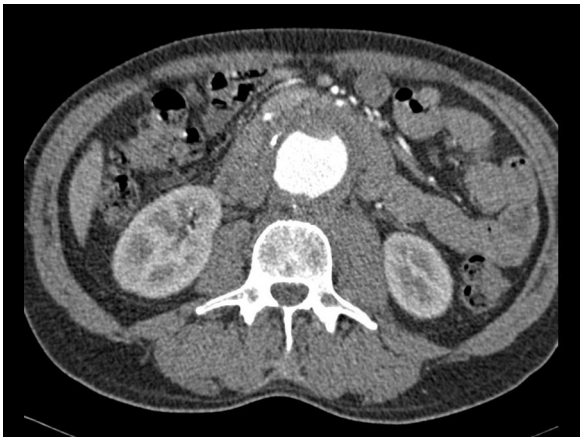

Fig. 1. Preoperative CT angiogram showing a 5.8 cm inflammatory abdominal aortic aneurysm.

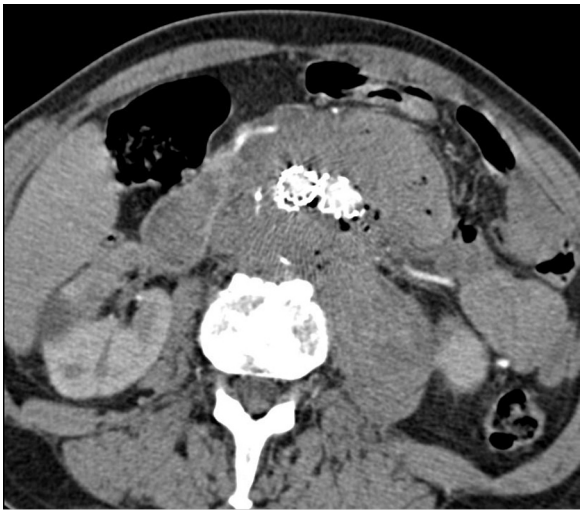

Fig. 2. CT angiogram of the aorta showing multiple bubbles of gas within the aneurysm sac surrounding the stent graft.

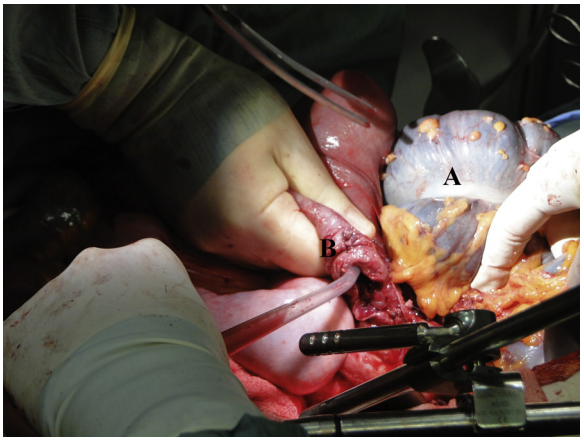

Fig. 3. Operative view of the duodenal defect.

the source. An enterotomy was done with retrograde passage of the enteroscope that detected a defect in the 3rd part of the duodenum as the source of the bleeding. Consequently, surgical exposure of the duodenal defect was done (Fig. 3), after which dissection of the abdominal aorta and identification of the fistula (Fig. 4) was achieved.

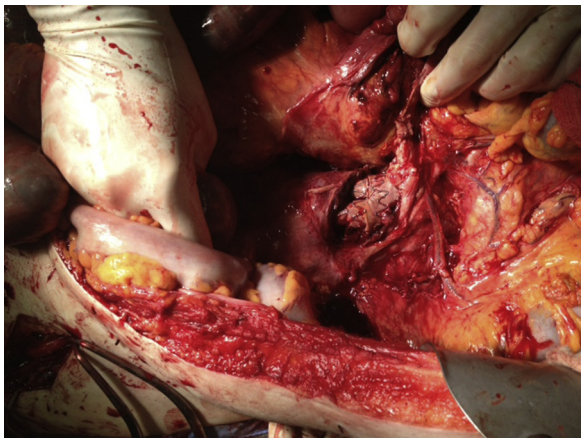

Fig. 4. Operative view of the defect in the AAA wall with exposure of the endograft.

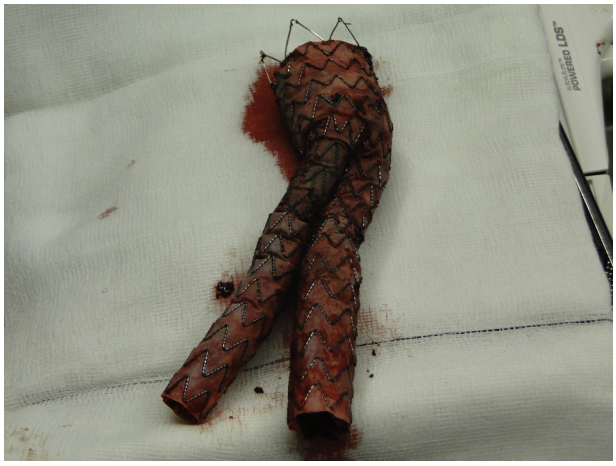

Fig. 5. Ex-vivo image of the endograft.

This was followed by surgical excision of the infected endograft (Fig. 5) with suturing of the proximal infrarenal aortic stump and distal iliac ends. The infected aortic sac was debrided and samples taken for pathological and microbiological analysis, which later proved out to be *Streptococcus anginosus*. Finally, trimming of the edges of the duodenal defect with surgical repair via a doudenorraphy was done.

Owing to the presence of extensive infection, and the hemodynamic instability of the patient, a decision was made to ensure haemostasis, after which part of the greater omentum was mobilized through a window in the transverse mesocolon which was stapled to close the aneurysm sac preventing further adhesions with the duodenal wall. The abdomen was closed and we started preparing the patient for an axillo-bifemoral bypass; however, owing to the critical condition of the patient, haemodynamic instability as well as upon anesthetics team requests, we decided to defer the axillo-bifemoral bypass to a second session after adequate resuscitation and stabilization. Unfortunately, as soon as the abdomen was closed, the patient went into ventricular fibrillation and all trials of resuscitation failed.

2. Discussion

The incidence of aortoenteric and aortocaval fistulae is higher in inflammatory abdominal aortic aneurysm than in atherosclerotic abdominal aortic aneurysm, despite the higher incidence of rupture in atherosclerotic abdominal aortic aneurysm.[3] Moreover, the endovascular approach through EVAR for inflammatory abdominal

aortic aneurysms is becoming the first-line therapy as it leads to improvement of periaortic inflammation.[4] In addition; the endovascular stent graft abolishes the contact between the suture line and the duodenum which was thought to play a major role in the development of aortoenteric fistulas following open repair of AAA.[5] Aortoenteric fistula after endovascular repair of abdominal aortic aneurysm occurs in approximately 0.36% of cases.[6] Such a complication has seldom been reported in literature following repair of an inflammatory abdominal aortic aneurysm, and this was attributed to the presence of thick aneurysm wall of inflamed tissues intervening between the aneurysm and surrounding structures.

Aortoenteric fistula represents one means of endograft infection, with other causes including skin contamination from interventional procedures, hematogenous graft seeding, and bacteremia. Secondary aortoenteric fistula is a well described complication of infrarenal abdominal aortic aneurysm repair. These fistulas typically present with signs of gastrointestinal bleeding, known as herald bleeding. Less frequently, these fistulas may present simply with signs of sepsis.[7] Parry et al. reported the first case of inflammatory AAA treated by EVAR who developed aortoduodenal fistula 7 months later, presenting with a picture of sepsis.[8] An argument could be made however to the accuracy of the decision to proceed with an EVAR in presence of increased inflammatory markers and suspicion of a pre-existing aortoenteric fistula as disclosed by the authors, and to this issue's contribution to the development of the fistula later on.

The definite aetiology of the development of aortoduodenal fistula in endovascularly managed inflammatory AAA is still not entirely understood. The most incriminated cause is infection, which may be spreading to the endograft either directly from a nearby source of infection, or haematologically from a distant source. Hausegger et al as well as Janne d'Othee et al both reported cases with other possible causes such as stent migration and kinking.[9,10] Another hypothesis attributes fistula formation to a reduction of the peri-aortic inflammatory mass after EVAR which brings the stent graft in proximity with the duodenum. There may be some merit in this postulation, especially since as many as 79% of cases with inflammatory AAA are adherent to the duodenum.[8] Moreover Ratchford et al. discussed the involvement of endoleaks, especially type I endoleaks in the development of aortoduodenal fistula leading to failure of device attachment and aneurysm expansion with resultant bowel fistulation.[5] Likewise, type IV endoleaks were incriminated as one of the possible contributors to the development of the aortoduodenal fistula; especially with the initial types of endografts, as reported by Norgren et al.[11]

Another question that is raised is the use of steroids in the peri-operative management of inflammatory AAA with raised inflammatory markers. This may be a difficult decision due to uncertainties regarding the origin of inflammatory reaction (sepsis or non-specific peri-aortic inflammatory reaction) and the effect of steroids on the outcome. Parry et al. argued that in the presence of a nonspecific periaortic mass, the absolute aetiology of which is unclear, the use of prednisolone is misplaced.[8]

In our case, the source of endograft infection was most probably direct extension from the psoas abscess; which is supported by the fact that microbiological examination of the infected aneurysm wall revealed *S. anginosus*, a subgroup of viridians Streptococci. To our knowledge necrotizing fasciitis with *S. anginosus* endograft contamination has only been reported four times before.[12] These organisms are recognized as normal flora of the human oral cavity and gastrointestinal tract with the ability to cause abscesses and systemic infections. The unique characteristic of the *S. anginosus* group that sets these Streptococci apart from other pathogenic Streptococci, such as *Streptococcus pyogenes* and *Streptococcus agalactiae*, is their ability to cause abscesses.[13]

Traditionally, management of aorto-enteric fistulas has consisted of extra-anatomic bypass combined with aortic ligation or in situ aortic reconstruction using femoral–popliteal vein graft, or cryopreserved graft, in conjunction with removal of the infected graft and duodenal reconstruction or repair.[14] Most publications are single case reports, but a meta-analysis yielded a 35% mortality.[1] Some authors prefer a single session repair, while others recommend a staged procedure with an initial extra anatomical axillo-bifemoral bypass followed by ligation of the aorta and iliacs.[15]

3. Conclusion

Aortoduodenal fistula is a rare but dangerous complication of EVAR, secondary to infection or endoleak. The inflammatory nature of the aneurysm does not protect the patient from such a complication, despite the documented decrease in inflammatory process surrounding the aneurysm following aneurysm exclusion via the endograft. Prompt diagnosis and intervention are crucial to avoid a fatal outcome. The major cause of such a complication is either infection or endoleaks. There is no time limit for the fistula to occur, and adequate protection against endograft infection must always take place. In addition, thorough preoperative exclusion of potential sources of graft infection is mandatory. Management of aortoduodenal fistulas is usually through primary or staged ligation of the abdominal aorta and iliacs in addition to axillobifemoral extra-anatomical bypass or aortic tube grafting according to the etiology and extent of aneurysm wall infection. Although early diagnosis and intervention increases chances of survival, this condition is usually fatal and carries a very grave prognosis; as in the unfortunate case of our patient.

Conflict of interest

None.

Funding

None.

Ethical approval

Written informed consent was obtained from the patient's next of kin for 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 contributions

Mr Mohamed Zaki: Data collection, data analysis and writing.
Mr Wael A.Tawfick: Data collection, data analysis and revision of manuscript.
Mr Mahmoud Alawy: Data collection, data analysis and writing.
Dr Mohammed ElKassaby: Data collection, data analysis and writing.
Ms Niamh M Hynes: Revision of manuscript.
Mr Sherif Sultan: Study design, revision of manuscript and final approval of manuscript.

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