

**AORTOENTERIC FISTULA, A COMPLICATION OF AORTIC ANEURYSM REPAIR –
AN INTERESTING CASE*****Saman Anwar, Ayesha Shayan, Rida Zainab, Ramsha Fatima, Hina Naseer, Jawaid Iqbal**

Department of Radiology, Liaquat National Hospital, and Medical College, Karachi, Sindh, Pakistan.

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Corresponding Author*Saman Anwar**Department of Radiology,
Liaquat National Hospital,
and Medical College,
Karachi, Sindh, Pakistan.**ABSTRACT**

Aortoenteric fistula (AEF) is an abnormal connection between the aorta or its major arterial branches and part of the gastrointestinal tract. It is fairly uncommon, but a rather fatal condition. It has either a primary or secondary etiology, secondary being more common. We report the case of a 61 year old woman with a history of hypertension who presented in the ER with lower abdominal pain and vomiting for the past 10 days. She underwent CTA of thoracoabdominal aorta and was diagnosed as abdominal aortic aneurysm, for which she later underwent surgery. About 45 days later, she was brought to the ER again with hematemesis and melena. Her CTA of thoracoabdominal aorta again showed abdominal aortic aneurysm adherent with adjacent small bowel loops with surrounding multiple air specks and extravasation of contrast from aortic to duodenal lumen, likely representing aortoenteric fistula. Esophagogastroduodenoscopy was done to secure bleeding site however due to poor hemodynamic status of patient, procedure was abandoned and a second surgery was performed day after, where fistulous communication was intervened and jejunostomy was made. 1 week later, the patient died due to sudden myocardial infarction. Our report highlights a fatal manifestation of aortoenteric fistula, and reviews the associated literature.

KEYWORDS: Aortoenteric fistula, aortic aneurysm, complication, gastrointestinal bleed.**INTRODUCTION**

Aortoenteric fistula is a rare cause of massive gastrointestinal bleed with a high degree of mortality. Aortic fistulas may be considered primary (associated with a complicated abdominal aortic aneurysm) or secondary (associated with graft repair). It needs prompt diagnosis with appropriate diagnostic modalities in order to preserve life.

CASE PRESENTATION

We report the case of a 61 year old woman with a history of hypertension who presented in the ER with lower abdominal pain and vomiting for the past 10 days. On an ultrasound from a peripheral hospital the query of abdominal aortic aneurysm was raised.

CTA of thoracoabdominal aorta (Figure 1a,b,c,d) was proceeded which showed saccular aneurysmal dilatation of infra renal part of abdominal aorta extending over a length of 9.3cm up to the origin of iliac arteries (Figure 1c) and its maximum diameter measured 9.5cm. It was thrombosed along its periphery and was causing compression and forward displacement of adjacent small bowel loops (Figure 1a, 1d).



Figure 1a

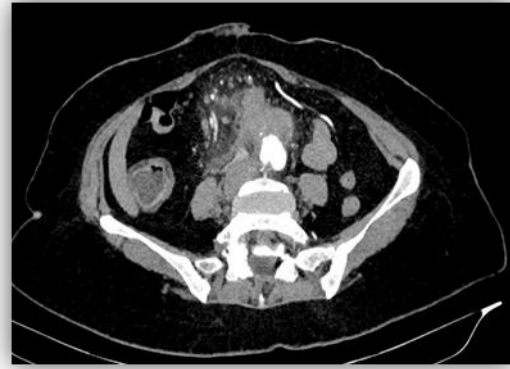


Figure 1b



Figure 1c



Figure 1d

Based on this CTA, diagnosis of infra renal abdominal aortic aneurysm was made. Patient was admitted under care of vascular surgery team and surgical intervention planned.

After written and informed consent, baseline investigations, pre-operative workup and general anesthesia fitness she underwent surgery. Perioperatively, the aneurysmal sac was identified to be adherent to D3 and D4 part of duodenum. During an attempt to separate them, there was a serosal tear in D4 segment of duodenum. However, it was repaired and aortic aneurysm graft repair was also done. The patient went through the smooth recovery with no drastic or minor complications in hospital post-operative course and was stable on the weekly follow-ups.

The patient presented again in the ER after 45 days of the surgery with complain of hematemesis and melena, and CTA was repeated which showed high density in bowel loops on plain images representing blood (Figure 2a, b). There was redemonstration of aneurysmal dilatation of infra renal part of abdominal aorta extending up to the origin of iliac arteries with postsurgical changes in it. It again appeared thrombosed along its periphery and adherent to adjacent small bowel loops with multiple surrounding air specks (Figure 2c). Extravasation of

contrast was seen from aorta to bowel lumen, likely representing aortoenteric fistula.



Figure 2a



Figure 2b



Figure 2c

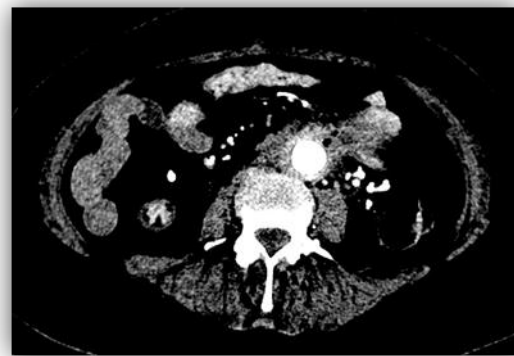


Figure 2d

Keeping in view the findings of CTA and patient's poor hemodynamic status, gastroenterology team was taken on board and esophagogastroduodenoscopy was done to secure hemostasis where the source of bleeding was identified at the level of D4 part of duodenum and in jejunum near the proximal suture line. Packing was done and the procedure abandoned due to faltering hemostasis. A second surgery was performed next day after the patient was stabilized, fistulous communication was intervened, jejunostomy was made and the vascular graft was found to be in optimal condition on assessment. Again the postoperative course went smooth and patient was stabilized day after the surgery. However after 1 week, she succumbed to death secondary to sudden myocardial infarction.

DISCUSSION

Aortoenteric fistula (AEF) is an abnormal connection between the aorta or its major arterial branches and part of the gastrointestinal tract. It is fairly uncommon with an incidence rate of 1.6 to 4%, but a rather fatal condition with a mortality rate of 24 to 45.8%.^[1]

In primary cases, the underlying etiology is most commonly aneurysmal development which results in

thinning of the walls of aorta and outermost layer of bowel. With each cardiac pulsation and also peristalsis, there is mechanical friction.^[2] Foreign bodies, tumors, inflammation, mycotic aneurysms, sepsis, syphilis, salmonella, and tuberculosis also exacerbate the erosion.^[3,4,5,6]

While secondary etiologies are more common, they follow after an open or endovascular surgical intervention on the aorta such as elective abdominal aortic aneurysm resection, aortic replacement or bypass for aorto-iliac occlusive disease, resection of ruptured aneurysm, and stent graft placement for aortic aneurysm.^[7,8] Mechanical rubbing of the graft against approximately lying bowel causes erosion into the enteral wall.^[9] The pathophysiology of secondary fistulas is multifactorial and related to foreign body reaction to the graft, graft kinking, superimposed infection, endoleak coil placement, and endotension.^[10,11]

In both primary and secondary aortoenteric fistula, the duodenum is the portion of the gastrointestinal tract most frequently involved, especially in the third section where the duodenum is in the closest proximity to the aorta.^[12]

The patients present with symptoms like pain related to the location of the lesion such as abdominal pain, back pain, or suprapubic pain, gastrointestinal bleeding seen as hematochezia, hematemesis, or melena, hemorrhagic shock, syncope, weight loss, anorexia, moderate to high fever, nausea and vomiting.^[13] Of these, the most common presentation is with melena and hemorrhagic shock, followed by abdominal pain and hematemesis.^[14]

Diagnostic workup may include esophagogastroduodenoscopy, however its sensitivity is only about 50%.^[15] CTA has widely variable sensitivity (40%–90%) and specificity (33%–100%) for the diagnosis of aortoenteric fistulas.^[16] CT findings in AEF include ectopic gas either within or directly adjacent to the aortic lumen or the graft, bowel wall thickening adjacent to the graft, effacement of the periaortic and perigraft fat plane, and loss of fat plane between the aorta and bowel, perigraft soft tissue, perigraft fluid, perigraft hematoma and dystrophic vascular graft calcification. The extravasation of contrast from the aorta into the bowel lumen is a highly specific sign, but extremely rare.^[16,17]

The optimal outcome of AEF requires confirmation of the diagnosis, quick control of the hemorrhage, repair of the bowel defect, eradication of associated infection, and revascularization. The only curative treatment for AEF is surgery, either with the open approach or endovascular, without which the mortality approaches 100%. While in-hospital mortality of patients undergoing open surgery is significantly higher than endovascular aortic, that is 33.9%. However, rate of late sepsis was higher in endovascular approach, where open surgery was associated with a 19% as compared to 42% in endovascular approach. Despite these differences, the survival rate between the techniques is similar.^[18]

It is all the more important to be vigilant in preventing the formation of an AEF. Male patients over the age of 65, especially those who have a smoking history, should undergo screening ultrasounds. Furthermore, during open surgical repair of abdominal aortic aneurysm the surgeon must ensure that the graft is sufficiently covered with a layer of tissue preventing the direct effect of mechanical friction with the adjacent structures. Approximating the aneurysmal sac around the graft is the common method. However omental flap or vascularized pedicle may also be used.^[19]

CONCLUSION

Although rare, an aortoenteric fistula has rather fatal consequences if not diagnosed and treated in a timely manner. Hence it is important that the clinician is well aware of the patient's history, especially pertaining to smoking, the common clinical presentations, and is able to opt for appropriate diagnostic tools. Symptoms like massive gastrointestinal bleed, hypervolemia and septicemia raise the suspicion of an aortoenteric fistula. CT scan is the modality of choice. The third segment of

the duodenum is most commonly involved segment. Prompt management is crucial and includes urgent surgery and antibiotics.

ETHICAL REVIEW

This case report was written and radiological images, surgical findings and other details were added after taking consent from patient's attendant. Patient's identity is not shown here and will be kept confidential.

CONFLICT OF INTEREST

The authors declared, there is no conflict of interest.

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REFERENCES

1. Saito, H., Nishikawa, Y., Akahira, Ji. *et al.* Secondary aortoenteric fistula possibly associated with continuous physical stimulation: a case report and review of the literature. *J Med Case Reports*, 2019; 13: 61.
2. Malik MU, Ucbilek E, Sherwal AS. Critical gastrointestinal bleed due to secondary aortoenteric fistula. *J Community Hosp Intern Med Perspect*, 2015; 5(6): 29677
3. Skourtis G, Papacharalambous G, Makris S, Kafikis F, Kastrisios G, Goulas S, Antoniou I, Giannakakis S, Maltezos C. Primary aortoenteric fistula due to septic aortitis. *Ann Vasc Surg*, 2010 Aug; 24(6): 825.e7-11.
4. McIntyre KE, Malone JM, Richards E, Axline SG. Mycotic aortic pseudoaneurysm with aortoenteric fistula caused by *Arizona hinshawii*. *Surgery*, 1982 Feb; 91(2): 173-7.
5. Goldbaum TS, Lindsay J, Levy C, Silva CA. Tuberculous aortitis presenting with an aortoduodenal fistula: a case report. *Angiology*, 1986 Jul; 37(7): 519-23.
6. Keunen B, Houthoofd S, Daenens K, Hendriks J, Fourneau I. A Case of Primary Aortoenteric Fistula: Review of Therapeutic Challenges. *Ann Vasc Surg*, 2016 May; 33: 230.e5-230.e13.
7. D. Bergqvist and M. Björck, "Secondary arterioenteric fistulation—a systematic literature analysis," *European Journal of Vascular and Endovascular Surgery*, 2009; 37(1): 31–42.
8. S. E. Wilson, R. S. Bennion, A. I. Serota, and R. A. Williams, "Bacteriological implications in the pathogenesis of secondary aorto-enteric fistulas," *British Journal of Surgery*, 1982; 69(9): 545–548.
9. Khalaf C, Houlind KC. Case report: Primary aortosigmoid fistula - A rare cause of lower gastrointestinal bleeding. *Int J Surg Case Rep*, 2017; 40: 20-22.
10. Bertges DJ, Villella ER, Makaroun MS. Aortoenteric fistula due to endoleak coil

- embolization after endovascular AAA repair. *J Endovasc Ther*, 2003; 10(1): 130–135
11. Chenu C, Marcheix B, Barcelo C, Rousseau H. Aorto-enteric fistula after endovascular abdominal aortic aneurysm repair: case report and review. *Eur J Vasc Endovasc Surg*, 2009; 37(4): 401–406.
 12. Malik, M.U., Ucbilek, E. and Sherwal, A.S. “Critical gastrointestinal bleed due to secondary aortoenteric fistula,” *Journal of Community Hospital Internal Medicine Perspectives*, 2015; 5(6): 29677.
 13. Luo, J., Tang, W., Wang, M. *et al.* Case series of aortoenteric fistulas: a rare cause of gastrointestinal bleeding. *BMC Gastroenterol*, 2021; 21: 49.
 14. Voorhoeve R, Moll FL, de Letter JA, Bast TJ, Wester JP, Slee PH. Primary aortoenteric fistula: report of eight new cases and review of the literature. *Ann Vasc Surg*, 1996 Jan; 10(1): 40-8.
 15. Deijen CL, Smulders YM, Coveliers HME, Wisselink W, Rauwerda JA, Hoksbergen AWJ, The Importance of Early Diagnosis and Treatment of Patients with Aortoenteric Fistulas Presenting with Herald Bleeds. *Annals of vascular surgery*, 2016.
 16. Vu, Q.D. *et al.* “Aortoenteric fistulas: CT features and potential mimics,” *RadioGraphics*, 2009; 29(1): 197–209. Available at: <https://doi.org/10.1148/rg.291075185>.
 17. Hagspiel, K.D. *et al.* “Diagnosis of aortoenteric fistulas with CT angiography,” *Journal of Vascular and Interventional Radiology*, 2007; 18(4): 497–504.
 18. Kakkos SK, Bicknell CD, Tsolakis IA, Bergqvist D., Hellenic Co-operative Group on Aortic Surgery. Editor's Choice - Management of Secondary Aortoenteric and Other Abdominal Arterio-enteric Fistulas: A Review and Pooled Data Analysis. *Eur J Vasc Endovasc Surg*, 2016 Dec; 52(6): 770-786
 19. Deriu GP, Ballotta E. Prevention of enteric erosion by vascular prostheses. *Tex Heart Inst J.*, 1982 Sep; 9(3): 325-8.