



GI Hemorrhage From an Arterio-Enteric Fistula From a Failed Pancreas Allograft

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ABSTRACT

Arterio-enteric fistulas involving pancreas allograft are rare complications of transplantation that manifest as gastrointestinal bleeding. We discuss the case of a 34-year-old patient with failed pancreas transplant who presented with recurrent sentinel bleeding before catastrophic hemorrhage. Multiple endoscopies did not identify the source of bleeding, and subsequent angiography demonstrated a fistulous connection between the transplanted pancreatic artery and the small bowel. A stent graft was placed with immediate stabilization. We provide a review of the literature and discuss the clinical manifestations, diagnosis, and management of arterio-enteric fistulas to highlight the importance of early recognition and intervention in preventing life-threatening bleeding.

INTRODUCTION

An arterio-enteric fistula (AEF) is a pathologic communication between the aorta and the gastrointestinal (GI) tract. Primary AEFs form when native aorta compresses and erodes into part of the GI tract, whereas secondary AEFs arise after surgical procedures involving the aortoiliac tree. The incidences of primary and secondary AEFs have been reported to range from 0.02% to 0.07% and 1%, respectively.¹ An AEF usually occurs in the setting of at least 1 of the following risk factors: atherosclerotic disease, hypertension, connective tissue disease, and history of aortic interventions.² AEFs will typically present with an initial minor, intermittent, “herald” bleed, followed by massive GI hemorrhage. AEFs in patients with pancreas allografts are rare complications of transplantation but are responsible for half of reported GI bleeding (GIB) episodes in this population, followed by anastomotic ulcer, donor duodenal bleed, pseudoaneurysms, and other rare causes such as mucosal rejection or cytomegalovirus-related ulcers.³ We present a case of GI hemorrhage from an AEF in a patient with a failed pancreas transplant.

CASE REPORT

A 34-year-old woman presented with severe epigastric pain and hematochezia. Her medical history was significant for hypertension and type 1 diabetes complicated by end-stage renal disease for which she received a simultaneous kidney and pancreas transplant with enteric exocrine drainage 4 years earlier. The kidney and pancreas allografts failed 1 year earlier because of chronic rejection and occlusive thrombus of the transplant pancreatic artery that required anticoagulation. She continued low-dose tacrolimus after transplant failure. On presentation, she was hypotensive and had a hemoglobin of 6.8 g/dL. She had no prior endoscopies. The initial esophagogastroduodenoscopy (EGD) was unremarkable and did not reveal a clear etiology; no blood or hematin was seen up to the second portion of the duodenum. While preparing for a colonoscopy, she had 4 large melanic bowel movements and subsequently became hypotensive and unresponsive. She was found to have a 3-g drop in hemoglobin. An emergent EGD showed a massive blood clot in the gastric body and fundus that was unsuccessfully removed; however, no fresh blood or active bleeding was identified. The following day, a repeat EGD was performed with adequate visualization of the entire stomach, revealing no obvious source for bleeding. Shortly thereafter, she developed massive hematochezia and hematemesis with significant hemodynamic instability requiring 4 vasopressors and 65 units of blood products. She was intubated for airway protection and was taken for an emergent arterial angiography, which demonstrated active hemorrhage into the small bowel through the transplant pancreatic artery (Figure 1). A stent

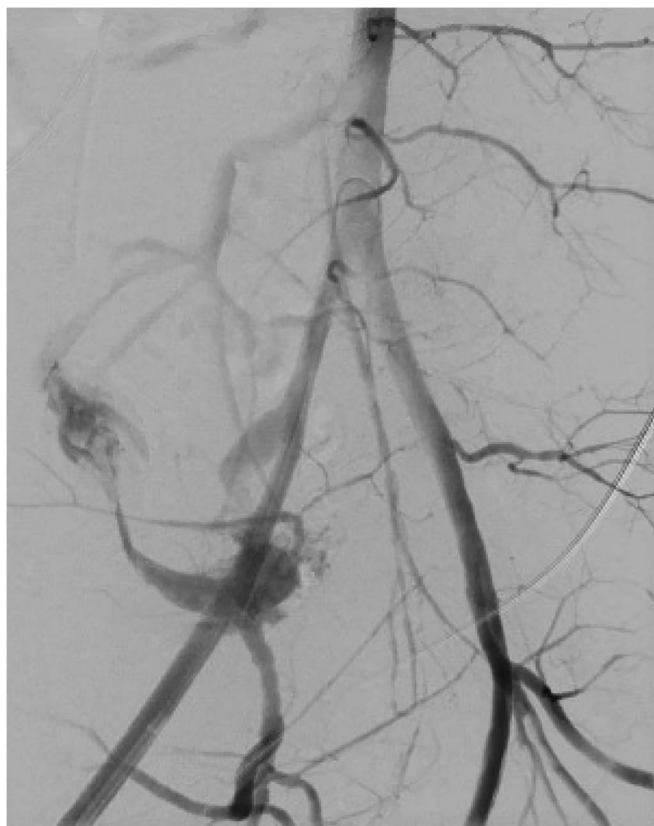


Figure 1. Angiogram showing contrast extravasation into the cavity and subsequently adjacent small bowel through the transplanted pancreatic artery from the right common iliac artery.

graft was placed at the right common iliac artery anastomosis site (Figure 2) and she immediately stabilized. A small bowel enteroscopy the next day showed allograft erosion into proximal jejunum with visible suture material (Figure 3). She was discharged home the following week without any further bleeding to date.

DISCUSSION

Pancreatic transplant with enteric drainage is the standard of care for patients with type 1 diabetes who develop end-stage renal disease. Transplanted pancreas blood supply is provided by the donor-to-recipient arterial anastomosis involving the iliac arterial system. Enteric drainage occurs by anastomosis between donor pancreas and portion of small bowel to recipient small bowel.⁴ Early complications immediately after transplantation include vascular thrombosis and allograft pancreatitis. Later complications, such as abscess or fistula formation, usually occur a few months after transplant.⁵ An AEF typically forms between the arterial Y graft and the donor duodenum with bleeding occurring through the small bowel anastomosis.⁶ AEF associated with pancreatic graft failure is extremely rare and described in a limited number of case reports. The pathophysiology is not well established but likely multifactorial. In the setting of graft failure, AEF formation may be because of a



Figure 2. CT reformat demonstrating the stent within the right common iliac artery. CT, computed tomography.

combination of chemical erosion from pancreatic enzymes and decreased vascular wall integrity due to immunosuppression and chronic rejection resulting in local inflammation.

To date, there are only 30 reported cases of AEF in patients with enteric-drained pancreas transplants, and the majority were associated with failed grafts.² The average time between graft failure and presentation was 10.4 months. The initial presentation in nearly all the cases was overt GIB, most commonly lower GIB.

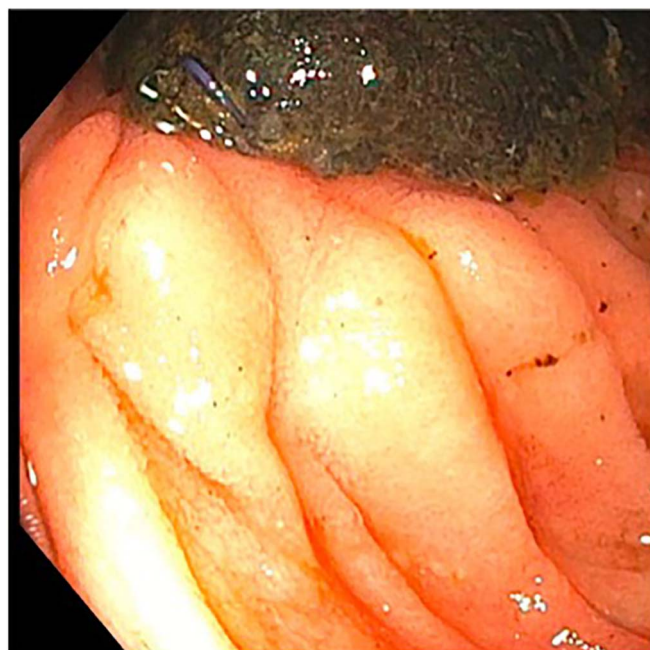


Figure 3. Proximal jejunum allograft site with visible suture.

Five patients died despite intervention in 4 of the cases. There were no cases that were diagnosed by endoscopic evaluation.

Although endoscopy is the gold standard for the evaluation of GIB, it only detects a quarter of secondary AEFs.^{7,8} Angiography is the preferred method for accurate diagnosis while definitive management includes endovascular stent or open surgery. Early recognition and appropriate intervention is crucial because GI hemorrhage among these patients carries a mortality rate of 100% if not definitively managed.⁹ AEFs can present with excessive bleeding making visualization difficult or as a sentinel bleed that is not active at the time of the endoscopy, as seen with our patient. Most AEFs also occur in the third or fourth portion of the duodenum which contributes to the variation in the diagnostic yield of endoscopy.¹⁰ Our report highlights that in a patient with failed pancreas transplant who presents with GI bleeding and inconclusive endoscopy, gastroenterologists should suspect AEF and act quickly to involve surgery or interventional radiology for definitive management.

AEF formation after chronic pancreatic graft failure is extremely rare and only reported in a limited number of case reports. Early angiography should be considered for localization and successful intervention. High suspicion and early recognition are key factors in preventing a later catastrophic hemorrhage.

DISCLOSURES

Author contributions: R. David wrote the article. J. Trieu and S. Shokoohi revised and edited the article. J. Trieu is the article guarantor.

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Informed consent was obtained for this case report.

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REFERENCES

1. Beuran M, Negoï I, Negoï RI, et al. Primary aortoduodenal fistula: First you should suspect it. *Braz J Cardiovasc Surg.* 2016;31(3):261–3.
2. Leiberman D, Sharma V, Siddagangiah V, et al. Radiological initial treatment of vascular catastrophes in pancreas transplantation: Review of current literature. *Transpl Rev.* 2021;35(3):100624.
3. Polanco N, Keihanian T, Barkin J, et al. Gastrointestinal bleeding after pancreatic transplant, beyond the usual suspects: A systematic review. In: Proceedings from the ACG 2019 Annual Scientific Meeting and Postgraduate Course; October 25–30, 2019; San Antonio, TX. Abstract S1550.
4. Hochberg C, Walker S, Applefeld W, et al. An unusual case of massive GI bleeding in a patient with pancreatic transplant. In: Proceedings from American Thoracic Society International Conference Abstracts; May 22, 2018; San Diego, CA. Abstract A5169.
5. McBeth BD, Stern SA. Lower gastrointestinal hemorrhage from an arterioenteric fistula in a pancreatorenal transplant patient. *Ann Emerg Med.* 2003;42(4):587–91.
6. Osband AJ, Laskow DA. Unusual source of gastrointestinal bleeding. *J Nat Sci.* 2015;1:e733.
7. Perler B, Miller S, Simons M, et al. Aortoenteric fistulas: A proposed stepwise diagnostic approach in an otherwise ambiguous clinical situation. In: Proceedings from the ACG 2018 Annual Scientific Meeting and Postgraduate Course; October 5–10, 2018; Philadelphia, PA. Abstract S1391.
8. Deijen CL, Smulders YM, Coveliers HME, et al. The importance of early diagnosis and treatment of patients with aortoenteric fistulas presenting with herald bleeds. *Ann Vasc Surg.* 2016;36:28–34.
9. Doherty B, Kilgore T, Pearson A. Aortoenteric fistula: A rare cause of gastrointestinal bleed requiring emergent surgical intervention. In: Proceedings from the ACG 2017 Annual Scientific Meeting and Postgraduate Course; October 13–18, 2017; Orlando, Florida. Abstract S1063.
10. Luo J, Tang W, Wang M, et al. Case series of aortoenteric fistulas: A rare cause of gastrointestinal bleeding. *BMC Gastroenterol.* 2021;21(49):49.

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