ACG CASE REPORTS JOURNAL

CASE REPORT | SMALL BOWEL

Splenic Pseudoaneursym as the Cause of Recurrent Gastrointestinal Bleeding in a Woman With Diffuse Scleroderma

Joshua Hartman, MD,[1]* Marion-Anna Protano, MD,[2]* and Barry Jaffin, MD[2]

[1]*Department of Internal Medicine, Mount Sinai Medical Center, New York, NY*
[2]*The Dr. Henry D. Janowitz Division of Gastroenterology, Mount Sinai Medical Center, New York, NY*

Abstract

A 67-year-old woman with a 15-year history of intestinal scleroderma presented with recurrent melena. Upper endoscopies revealed a healing, non-bleeding, large gastric ulcer. After the third bleed, angiography demonstrated bleeding from a splenic artery pseudoaneurysm adjacent to the gastric ulcer. Scleroderma patients are at risk of bleeding from esophagitis or gastric arteriovenous malformations, while splenic artery pseudoaneurysms are primarily attributed to pancreatitis and trauma. This is the first reported case of gastrointestinal bleeding from a splenic artery pseudoaneurysm in a patient with intestinal scleroderma and a large gastric ulcer.

Introduction

Scleroderma is a chronic autoimmune disease involving the microvascular system and leads to increased fibroblast activation and excessive production of collagen.[1] Vascular disease in scleroderma occurs in nearly every organ and is responsible for many manifestations of scleroderma, such as Raynaud's phenomenon.[1] Splenic artery pseudoaneurysms (SAPA) are extremely rare; to date, less than 200 cases have been reported in the literature.[2] SAPAs often present as massive, unstable hemorrhaging, typically from the pancreatic duct, and less commonly from the stomach and colon.[3-8]

Case Report

A 67-year-old woman presented with fatigue and melena. She had a past history of intestinal scleroderma and peripheral vascular disease. Her scleroderma was diagnosed 15 years earlier when she had a pseudo-obstruction requiring partial colectomy. Her medications included octreotide and a proton pump inhibitor; she was not taking non-steroidal anti-inflammatory drugs. Physical exam was significant for mild abdominal distention without tenderness and guaiac-positive brown stool. Initial labs revealed hemoglobin 6.8 g/dL. Esophagogastroduodenoscopy (EGD) showed one 4-cm deep, clean-based ulcer in the cardia with no visible vessel, which did not require endoscopic therapy (Figure 1). Biopsies showed chronic gastritis and ulceration. Immunohistochemical staining was negative for *H. pylori*. The patient was discharged 4 days later.

She returned to the emergency room 2 weeks later with weakness, melena, and hematemesis, and was found to be hypotensive and tachycardic with a hemoglobin of 5.3 g/dL. Repeat EGD showed numerous clots within the cardia and fundus of the stomach that could not be cleared and obscured vision of the gastric ulcer. CT angiography did not identify a bleeding site. Five days later, she was discharged with no further bleeding.

The patient returned to the emergency department for a third time 2 days later reporting numerous episodes of melena and hematemesis. Celiac angiography demonstrated a 6-mm distal splenic artery pseudoaneurysm,

ACG Case Rep J 2015;2(2):98-100. doi:10.14309/crj.2015.17. Published online: January 16, 2015.

Correspondence: Barry Jaffin, MD, Mount Sinai School of Medicine, 620 Columbus Avenue, New York, NY 10024 (Barry.Jaffin@mssm.edu).

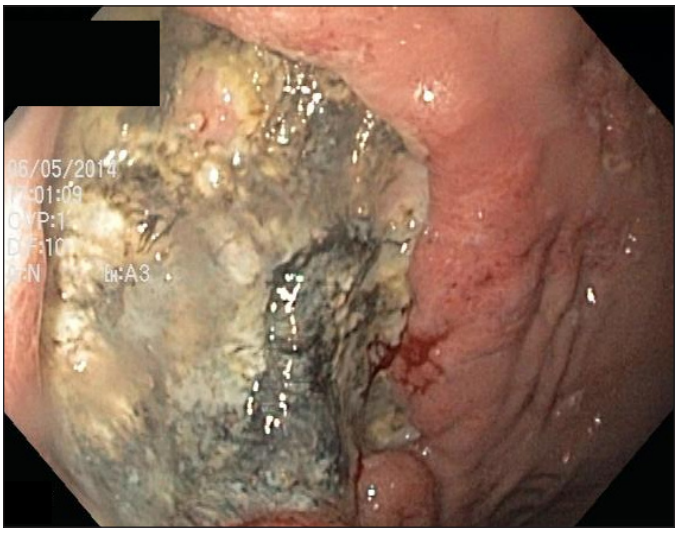

Figure 1. Clean-based gastric ulcer found via EGD on index bleed.

which communicated with the gastric lumen at the location of the prior ulcer (Figure 2). The pseudoaneurysm was embolized with n-BCA glue with resolution of active extravasation. The patient was discharged home 10 days later on pantoprazole. Repeat EGD 3 months later showed a healing, 1-cm gastric ulcer. She has been clinically stable for 4 months without any signs of gastrointestinal bleeding.

Discussion

The most common etiology of SAPA is pancreatitis (57%); other causes include trauma (27%), postoperative complications (2.7%), and peptic ulcer disease (2%).[2,9] Nearly half (47%) of SAPAs were identified in patients who presented with bleeding from the GI tract, including hematochezia, melena, and hematemesis.[2] In patients with pancreatitis-related SAPAs, digestion of the splenic artery by pancreatic enzymes may lead to weakening of arterial wall,[7] or presence of a longstanding pseudocyst may cause vascular erosion from enzymes within the pseudocyst and lead to direct compression or ischemia.[7] In traumatic SAPAs, rapid deceleration may lead to damage of the intima and elastic lamina of the splenic artery.[3] The relationship between SAPAs and gastric ulcers are less clearly defined; we postulate that ulcers may cause focal inflammation that can lead to SAPA.

Previous case reports exist of aneurysms in patients with scleroderma.[1,10] Macrovascular disease in scleroderma has been attributed to decreased vessel elasticity and microangiopathy of vasa vasora causing vascular wall ischemia.[1] The development of SAPA in scleroderma may have a similar mechanism due to the changes in the vessel walls. Scleroderma patients are at risk for watermelon stomach, telangiectasias, angiodysplasia, and esophagitis as possible causes of gastrointestinal bleeding.[11,12] There is no clear association between scleroderma and gastric ulcers, although there is 1 case report of a patient with peptic ulcers thought to be secondary to scleroderma.[13]

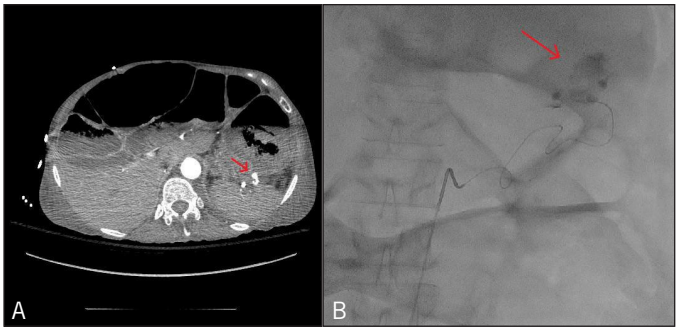

Figure 2. (A) CT angiography showing splenic pseudoaneurysm (SAPA), which required (B) glue embolization.

SAPA is often diagnosed by imaging with CT scan or ultrasound.[2,14] A review found the mean diameter of SAPAs to be 4.8 cm (range 0.3–17 cm),[2] thought smaller pseudoaneurysms are often missed.[2,14] Once identified, all SAPAs must be treated due to the high risk of rupture.[2] Size is not a determinant of rupture; in one review, the smallest (0.3 cm) and largest (17 cm) SAPAs were ruptured at presentation.[2] Angiography with transcatheter embolization has supplanted surgical treatment as it can be performed quickly and less invasively.[15]

We present the first report of a SAPA in a patient with scleroderma and a gastric ulcer with no history of pancreatitis. Our case highlights the importance of angiography in a patient with a repeat gastrointestinal bleeding from a large peptic ulcer. Pseudoaneurysms are a rare, but potentially fatal, cause of gastrointestinal bleeding and should be considered in the differential diagnosis.

Disclosures

Author contributions: J. Hartman and MA Protano wrote and approved the case report and reviewed the literature. B. Jaffin designed, revised, and approved the case report, and is the article guarantor. *Drs Hartman and Protano share first authorship.

Financial disclosure: None to report.

Informed consent was obtained for this case report.

Received: August 20, 2014; Accepted: December 10, 2014

References

1. Kaku Y, Kouda K, Yoshimura S, Sakai N. Cerebral aneurysms in scleroderma. *Cerebrovasc Dis*. 2004;17(4):339–41.
2. Tessier DJ, Stone WM, Fowl RJ, et al. Clinical features and management of splenic artery pseudoaneursym: Case series and cumulative review of literature. *J Vasc Surg*. 2003;38(5):969–74.
3. Piotrowska-Staworko G, Sek G, Kuklinski A, et al. Massive bleeding from the upper digestive tract in a patient with pseudoaneurysm of splenic artery. *Pol Merkur Lekarski*. 2006;20(120):698–701.
4. Wang J, Wang B, Chen DF. An unusual cause of recurrent massive upper gastrointestinal bleeding. *Gastroenterology*. 2012;143(3):542,872
5. Soga K, Konishi H, Miyajima T, et al. Bleeding from major and minor duodenal papillae due to hemosuccus pancreaticus confirmed with side-viewing gastroduodenal endoscopy. *Indian J Gastroenterol*. 2011;30(6):284.
6. Balsarkar DJ, Joshi MA. Rupture of splenic artery pseudoaneurysm presenting with massive upper gastrointestinal bleed. *Am J Surg*. 2002;183(2):197–8.
7. Walker TG, Geller SC, Waltman AC. Splenic artery pseudoaneurysms causing lower gastrointestinal hemorrhage. *Am J Roentgenol*. 1988;150(2):433–434.
8. Taslakian B, Khalife M, Faraj W, et al. Pancreatitis-associated pseudoaneurysm of the splenic artery presenting as lower gastrointestinal bleeding: Treatment with transcatheter embolisation. *BMJ Case Rep*. 2012;ii:bcr2012007403.
9. Norotsky MC, Rogers FB, Shackford SR. Delayed presentation of splenic artery pseudoaneursym following blunt abdominal trauma: Case reports. *J Trauma*. 1995;38(3):444–7.
10. Attaran RR, Guarraia D. Ascending aortic aneurysm in a man with scleroderma. *Clin Rheumatol*. 2007;26(6):1027–8.
11. Marie I, Antoniette M, Houivet E, et al. Gastrointestinal mucosal abnormalities using videocapsule endoscopy in systemic sclerosis. *Aliment Pharmacol Ther*. 2014;40(2):189–9.
12. Nishimagi E, Tochimoto A, Kawaguchi Y, et al. Characteristics of patients with early systemic sclerosis and severe gastrointestinal tract involvement. *J Rheumatol*. 2007;34(10):2050–5.
13. Platstira SC, Tzivras M, Vlachoyiannopoulous PG. Severe gastrointestinal involvement in systemic sclerosis. *Clin Rheumatol*. 2007;26(6):1025–6.
14. Savastano S, Feltrin GP, Antonio T, et al. Arterial complications of pancreatitis: Diagnostic and therapeutic role of radiology. *Pancreas*. 1993;8(6):687–92.
15. Xin, J, Xiao-Ping L, Wei G, et al. The endovascular management of splenic artery aneurysms and pseudoaneurysms. *Vascular*. 2011;19(5):257–61.