

CASE REPORT

Case report: An unusual case of cecal mucosal bleeding

Maseray S. Kamara  and Choichi Sugawa 

The Michael and Marian Ilitch Department of Surgery, Wayne State University, Detroit Medical Center, Detroit, Michigan, USA

Key words

apixaban, bleeding, cecal, gastrointestinal, mucosal, uremia.

Accepted for publication 15 September 2022.

Correspondence

Maseray S. Kamara, The Michael and Marian Ilitch Department of Surgery, Wayne State University, Detroit Medical Center, Suite 400, 3990 John R, Detroit, MI 48201, USA.
Email: maseraykam@gmail.com

Declaration of conflict of interest: All authors declare that they have no conflict of interests or disclosures regarding any potential benefits in any form from a commercial party related directly or indirectly to the subject of this manuscript.

Financial support: The author(s) received no financial support for the research, authorship, and/or publication of this article.

Introduction

Background. Acute gastrointestinal (GI) hemorrhage is a common clinical problem that can be broadly categorized by location as upper versus a lower GI bleed. Twenty to 30% of patients presenting with major gastrointestinal (GI) bleed will have a source distal to the ligament of Treitz and categorized as the latter. Lower GI bleeds typically arise from the colon, but other possibilities are within the small bowel. The differential diagnosis¹ of lower GI bleed commonly includes diverticulosis, arteriovenous malformations, ischemic colitis, colorectal neoplasms, radiation proctitis, anorectal disorders, and post-polypectomy bleeding.

Multidisciplinary management of lower GI bleeding is essential—emergency medicine, gastroenterology, surgery, interventional radiology, and intensivists all can play a crucial role. While the majority of gastrointestinal bleeding resolves spontaneously, approximately 15% of all gastrointestinal hemorrhages are cases of life-threatening bleeding resulting in the need of emergent resuscitation, diagnosis, and therapeutic intervention.^{1–3}

Case report

Presentation and initial assessment. This is a 73-year-old woman with end-stage renal disease and paroxysmal

Abstract

Cecal mucosal bleeding is an undocumented and rare cause of lower gastrointestinal (GI) bleeding. We present a case of a 73-year-old woman with end-stage renal disease and paroxysmal atrial fibrillation on apixaban who presented with lower gastrointestinal bleed. She was found to have symptomatic, acute chronic anemia requiring multiple packed red blood cell transfusions. Colonoscopy revealed a localized area of active, cecal mucosal bleeding without evidence of Dieulafoy lesion, ulcer, mass, arteriovenous malformation, or diverticula. Hemostasis was achieved with epinephrine injection and the use of bipolar electrocautery. She was later resumed on her therapeutic anticoagulation without recurrence of bleeding. Therapeutic anticoagulation in our patient with ESRD increased her risk for gastrointestinal bleeding. Had this transient, mucosal-limited bleeding not been active during endoscopic evaluation, the etiology of her massive gastrointestinal bleeding would have been missed. This case expands the differential of acute, lower GI bleeding to include cecal mucosal bleeding, which is a rare, intermittent, cause of bleeding that is amenable to endoscopic management.

atrial fibrillation on apixaban who presented after 3 days of having maroon stool, which progressed to hematochezia. On presentation, the patient endorsed weakness and fatigue, resulting in missed hemodialysis. She was normotensive with mild sinus tachycardia. On physical examination, her abdomen was soft, non-tender, and non-distended; her initial rectal examination was significant for maroon stool and external hemorrhoids. Laboratory studies demonstrated acute on chronic anemia (hemoglobin of 5.6 gm/dL) and electrolyte derangements reflective of missed hemodialysis.

Hospital course and clinical outcome. The patient was admitted and dialyzed. Bleeding progressed from maroon stools to frank hematochezia. She required 5 Units packed red blood cell transfusions, and ultimately underwent endoscopic evaluation on hospital Day 3. Colonoscopy revealed a localized area of active, mucosal bleeding at the cecum (Fig. 1a,b) without evidence of Dieulafoy lesion, ulcer, mass, arteriovenous malformation, or diverticula. Initial hemostasis was not successful with 10 cc of 1% normal saline-epinephrine solution (Fig. 1c). Hemostasis was ultimately achieved after application of bipolar electrocautery (Fig. 1d). She was later resumed on her therapeutic anticoagulation without recurrence of bleeding during her hospital stay and was discharged on hospital Day 4. Following

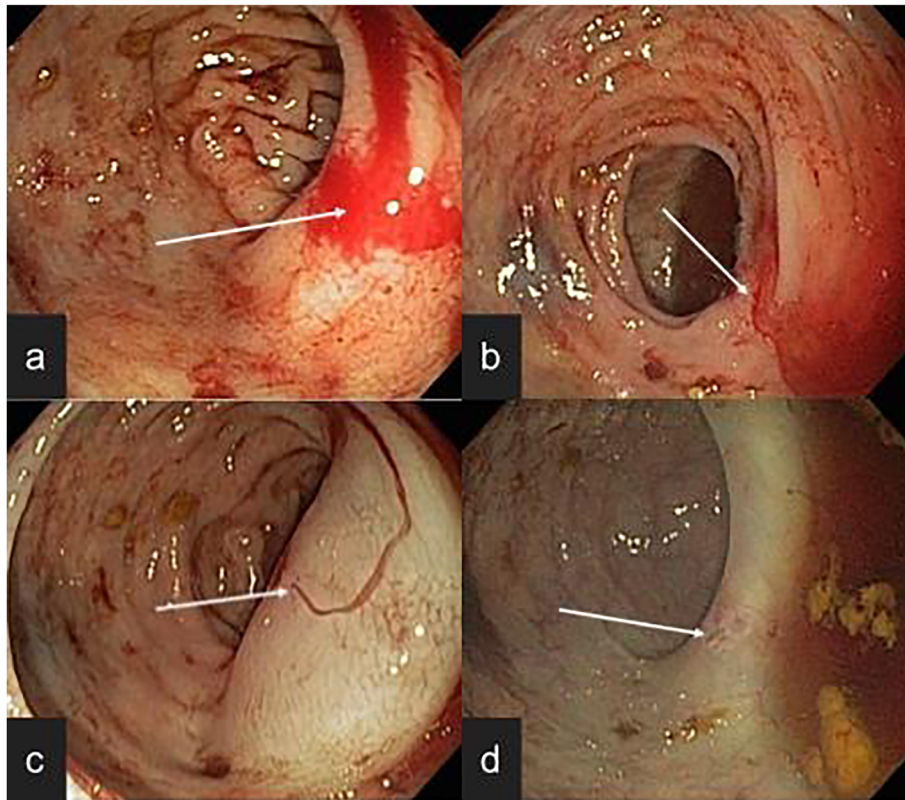


Figure 1 Diagnostic and after therapeutic intervention endoscopic findings. (a) Active cecal mucosal bleeding (arrow). (b) After irrigation, bleeding noted from an isolated, single spot (arrow). (c) After 10 cc of normal saline-epinephrine (1:10000) injection, bleeding persisted from the identified spot without apparent underlying lesion (arrow). (d) Hemostasis was achieved after a few applications of bipolar coagulation. No definite lesion can be seen except evidence of iatrogenic electrocautery (arrow).

discharge, the patient had no subsequent readmissions for rebleeding.

Discussion

Diagnosis. After identification of a gastrointestinal bleed, the key is to obtain a directed history and physical and begin initial resuscitation as indicated. The next step is the localization of acute hemorrhage. Upper endoscopy is critical in identifying a GI source proximal to the ligament of Treitz, but further a negative evaluation points to the lower GI tract as source. For the latter, colonoscopy is the diagnostic and therapeutic method of choice. If unable to be identified, the roles of tagged RBC, capsule endoscopy, and angiography come into play.¹⁻³

The bleeding debacle. The problem of transient or self-limiting or intermittent cause of bleeding can lead to diagnostic dilemmas that can greatly frustrate clinicians and patients alike. This case report focuses on cecal mucosal bleeding, which is an undocumented and rare cause of lower gastrointestinal bleeding (LGIB). In the identified case, the bleeding source was only identified because it was actively ongoing. The difficulty in diagnosis without gross physical lesions is important to establish in the

literature as a rare, but possible cause of the LGIB without an identifiable source.

This case expands the differential of massive lower GI bleeding to include cecal mucosal bleeding: a likely intermittent cause that is amenable to endoscopic management with epinephrine and electrocautery. While there have been reported cases of cecal Dieulafoy lesions⁴ and other submucosal masses,⁵ this endoscopic evaluation did not identify any visible, gross lesion as the cause for bleeding other than the mucosa. The patient's mucosal bleeding was identified in the setting of uremic platelet dysfunction³ and therapeutic anticoagulation (apixaban).

Uremic platelet dysfunction. The pathophysiology of platelet dysfunction of uremia is multifactorial with many mechanisms leading to platelet dysfunction and an abnormal platelet vessel wall interaction.⁶

The platelet count is usually normal to slightly low. In the setting of uremia, prostacyclin increases, which is a potent anti-aggregatory substance generated by the vessel wall. Further, in uremia, there is abnormal Von Willebrand factor (vWF), largest polymers are deficient, and there is defective interaction between vWF and glycoproteins; thus, reducing platelet ability to adhere to the endothelium and accomplish the hemostasis cascade.⁶

The interaction of these mechanisms leads to inhibition of hemostasis and risk for bleeding, which can result in gastrointestinal bleeding in uremic patients.

Gastrointestinal bleeding on apixaban. This patient was on apixaban, a highly selectively, reversible factor Xa inhibitor, for her atrial fibrillation. Compared with traditional, systemic anticoagulation, such as warfarin, NOACs (non-vitamin K antagonist oral anticoagulants), such as apixaban have been shown to have a more favorable safety profile; however, from multiple meta-analyses and phase IV studies, the risk of bleeding, particularly gastrointestinal bleeding (GIB), is still a concern in high-risk patients.

Based on a recent meta-analysis, which included 17 RCTs with over 75 000 patients, there is an increased risk of gastrointestinal bleeding among NOAC users; however, there is heterogeneity among different NOACs with both dabigatran and rivaroxaban (OR 1.58 and 1.48, respectively) associated with higher risk of GIB compared, less commonly seen with apixaban and edoxaban.⁷

Another point of consideration is that NOACs are renally excreted, thus patients with impaired renal function, such as this patient with ESRD on hemodialysis, are more likely to have drug accumulation, and hence a higher bleeding risk.

While being on anti-coagulation did increase the patient's risk of GI bleed, apixaban has been associated with less risk for bleeding compared with other NOACs.⁷

Clinical commentary. Mucosal bleeding is very difficult to identify with endoscopy. This patient was bleeding from a small area with an intermixed clot. After irrigation of the area, an active, pinpoint bleeding site was found. This is both a rare and unique finding. There has been a case reported of GIB secondary to spontaneous intramural intestinal hemorrhage on a patient on therapeutic anticoagulation.⁸ Such rare, spontaneous bleeding is associated with an abnormal coagulation profile, which can be seen in patients on therapeutic anticoagulation. This cited case report goes further to correlate nonspecific sonographic findings with the identified intramural intestinal hemorrhage.⁸ Further, the author (C.S), who performed surgical endoscopy for over 50 years at a busy urban, acute care hospital, experienced only two other cases of isolated mucosal bleeding (one from fundus of the stomach and one from posterior wall of the duodenal bulb) out of more than 50 000 GI bleeding patients. These cases were

able to be identified after irrigation revealed normal mucosa (without evidence of ulceration, mass, etc.) that was actively bleeding. Thus, mucosal bleeding is an underidentified and rare etiology of gastrointestinal bleeding.

Conclusion

In this case, massive LGIB was secondary to cecal mucosal bleeding in the setting of a patient on therapeutic anticoagulation, and additionally, ESRD with associated uremic platelet dysfunction. Such intestinal mucosal bleeding that was identified in this case is a clinically significant and rare cause of gastrointestinal hemorrhage. Diagnosis is further made difficult as the bleeding must be actively ongoing at the time of endoscopic evaluation to be diagnosed. Due to the intermittent nature of this type of bleeding, it is important to include cecal mucosal bleeding as a possible cause of obscure gastrointestinal bleeding, which is defined as bleeding of uncertain cause after a nondiagnostic upper endoscopy, colonoscopy, and barium small bowel follow-through.⁴

References

- 1 Tavakkoli A, Ashley S. Acute Gastrointestinal Hemorrhage. In: Townsend Jr CM, Beauchamp RD, Evers BM, Mattox MD (eds). *Sabiston Textbook of Surgery*. 20th ed. Philadelphia, PA: Elsevier - Health Sciences Division, 2016;1160–81.
- 2 Lawrence S, Friedman LS. Obscure Overt GI Bleeding. In: Feldman M, Friedman LS, Brandt LJ (eds). *Sleisenger & Fordtran's GI and Liver Disease*, 10th edn. Philadelphia: Saunders Elsevier, 2016; 328–33.
- 3 Muftah M, MulKi R, Dhare T, Chawla S KS. Diagnostic and therapeutic considerations for obscure gastrointestinal bleeding in patients with chronic kidney disease. *Ann. Gastroenterol.* 2019; **2019**: 113–23.
- 4 Dailey J, Russell MB, Sterling M. An Unusual Cause of Lower Gastrointestinal Bleeding: Cecal Dieulafoy's Lesion. *Cureus.* 2020; **12**: e7928.
- 5 Beck M, He M. Bleeding caecal mass: a rare finding. *J. Surg. Case Rep.* 2015; **9**: 1–3.
- 6 Salman Ş. Uremic Bleeding: A Concise Review. *Tepecik Egit Hast Derg.* 1994; **4**: 7–13.
- 7 Cheung KS, Leung WK. Gastrointestinal bleeding in patients on novel oral anticoagulants: Risk, prevention and management. *World J. Gastroenterol.* 2017; **23**: 1954–63.
- 8 Ribeiro H, Azevedo R, Pereira E, Banhudo A. Spontaneous intramural intestinal hemorrhage due to anticoagulation therapy. *Rev. Esp. Enferm. Dig.* 2018; **110**: 123.