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Single Case

Gastrointestinal Bleeding from Dieulafoy's Lesion in the Cecum

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Abstract
Dieulafoy's lesion is a rare cause of gastrointestinal (GI) bleeding comprising approximately 2% of all acute GI bleeds. It is an abnormal submucosal artery that has a tortuous course before protruding through the mucosa and leading to hemorrhage. Dieulafoy's lesions are most commonly located in the upper GI tract within the lesser curvature of the stomach. Lower GI tract Dieulafoy's lesions are remarkably rare. Our case describes an elderly gentleman who presented with fatigue and dyspnea several days prior to experiencing any evidence of GI bleeding. Initial laboratory investigation revealed severe anemia, requiring packed red blood cell transfusion. Endoscopic examination revealed a cecal Dieulafoy's lesion with active spurting of blood. Hemostasis was achieved through local epinephrine injection and hemostatic clipping. Previously reported cases of cecal Dieulafoy's lesions involve variable presentations including hematochezia, melena, or bright red blood per rectum. These lesions can be treated by angiography, surgically, or endoscopically via techniques that include epinephrine or ethanol injection, argon plasma coagulation, heater probe coagulation, hemostatic clips, or band ligation. Dieulafoy's lesions of the lower GI tract should be considered when no clear culprit is discovered, particularly with the concomitant use of antiplatelet agents and anticoagulants.

Introduction

Dieulafoy's lesion is a rare cause of gastrointestinal (GI) bleeding comprising approximately 2% of all acute GI bleeds [1, 2]. It is defined as a wide diameter (1–3 mm) tortuous artery that runs within the submucosal layer of the GI tract and eventually protrudes through

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the mucosa, causing hemorrhage [3]. Disease presentation typically involves painless, intermittent, and recurrent bleeding [4]. The vast majority of Dieulafoy's lesions are located within the upper GI tract, particularly at the lesser curvature of the stomach within 10 cm of the gastroesophageal junction, followed by the duodenum and esophagus [2]. Dieulafoy's lesions within the lower GI tract are exceedingly rare and comprise approximately 5% of total cases [2]. Considering disease epidemiology, GI Dieulafoy's lesions are more often present in males as compared to females (2:1 ratio) and usually occur after the 5th decade of life [5]. We hereby present the case of a patient with lower GI bleeding caused by a Dieulafoy's lesion identified in the cecum and compare it with previously reported similar cases.

Case Report

An 81-year-old gentleman presented to the emergency department with a 3-day history of exertional dyspnea associated with excessive fatigue. He has been recently diagnosed with atrial fibrillation and started on apixaban 1 month earlier. No reports of melena, hematochezia, or bright red blood per rectum (BRBPR). Past medical history includes hypertension and aortic regurgitation with surgical bioprosthetic aortic valve replacement 10 years prior. Home medications include apixaban 5 mg twice daily. There was no history of cigarette smoking, nonsteroidal anti-inflammatory drugs, or recreational drug use. He reports no personal history of GI bleeding but endorses a family history of GI bleeding in his brother and father of unknown etiology. Physical exam revealed a pale, lethargic gentleman with a normal abdomen and rectum. There was no evidence of melena, hematochezia, BRBPR, or a rectal mass. Initial laboratory investigation revealed: hemoglobin 6.6 g/dL, hematocrit 20%, mean corpuscular volume 107 fL, platelet count 129,000/μL, ferritin 14 ng/mL, iron 44 μg/dL, iron-binding capacity 315 μg/dL, and vitamin B12 178 pg/mL. Two units of packed red blood cells were transfused alongside intravenous ferrous gluconate and intramuscular cyanocobalamin. Hospital course was complicated with two bouts of severe hematochezia. Urgent computed tomography angiography of the abdomen and pelvis did not reveal an active source of bleeding (shown in Fig. 1a, b). Emergent esophagogastroduodenoscopy the following morning was unrevealing. A colonoscopy promptly followed and revealed blood throughout the entire colon with fresh red blood concentrated within the ascending colon. An actively bleeding Dieulafoy's lesion was located in the cecum (shown in Fig. 2a, b). Two milliliters of 1:10,000 epinephrine solution were injected, and 5 hemostatic clips were deployed, achieving hemostasis (shown in Fig. 3). Post-procedure course remained stable with no recurrence of hematochezia. His fatigue abated,

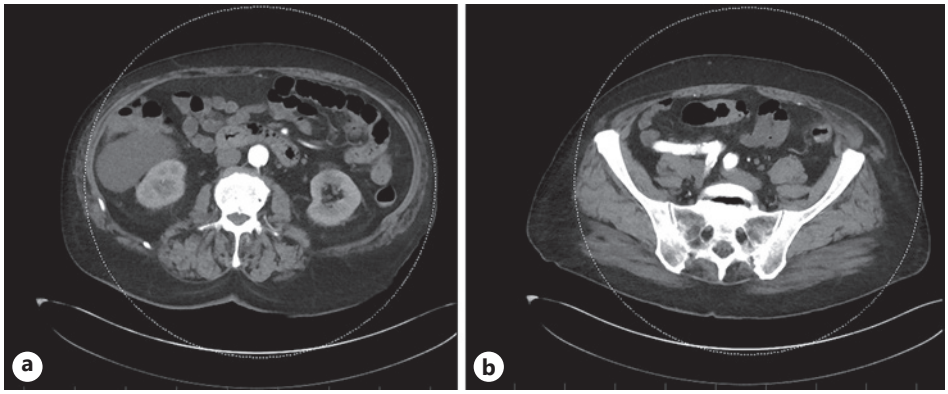

Fig. 1. a, **b** Computed tomography image revealing colonic stool with no active GI bleeding.

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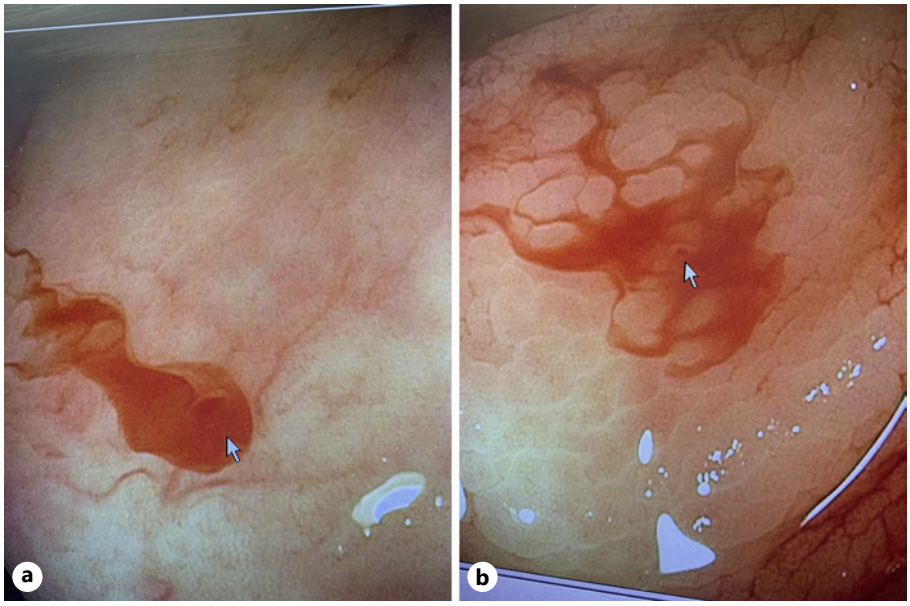

Fig. 2. a Active GI bleeding (white arrow) noted within the cecum. **b** Active arterial bleed (white arrow) within the cecum.

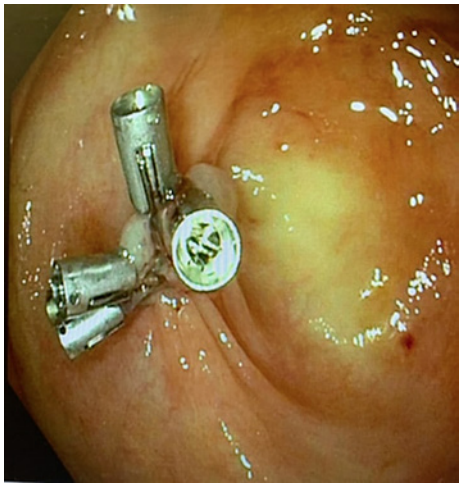

Fig. 3. Deployment of 5 hemostatic endoscopic clips with successful achievement of hemostasis.

and he was eventually discharged 48 h later in stable condition. Anticoagulation was resumed with no further fatigue or GI bleeding.

Discussion

Dieulafoy's lesions are a rare cause of acute GI bleeding and, when present, are most often located in the upper GI tract [2]. Their association with lower GI bleeding is exceedingly rare [2]. Other more common causes of lower GI bleeding include colonic diverticulosis, angiodysplasia, ischemic colitis, infectious colitis, neoplasia, hemorrhoids, and inflammatory bowel disease [6]. Diverticular disease is the most common cause of hematochezia, comprising 17–40% of cases with vascular ectasias (2–30%) and colitis (9–21%) following in second and third place [6].

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Table 1. Description of previously reported GI hemorrhage caused by Dieulafoy's lesions within the cecum

Presenting symptom	Age, years	Gender	NSAID/antiplatelet	Anticoagulation	Treatment modality	Reference
Hematochezia	25	Male	No	No	Right hemicolectomy	Pishori et al. [15]
Hematochezia	82	Female	Yes	Unknown	Hemostatic clip	Kinoshita et al. [12]
Hematochezia	34	Male	Unknown	Unknown	Hemostatic clip and ethanol	Sone et al. [10]
Hematochezia	39	Male	Unknown	Unknown	Hemostatic clip	Fukita [11]
Hematochezia	62	Female	Unknown	Unknown	Epinephrine and thermocoagulation	Singh et al. [13]
BRBPR	64	Female	Unknown	Unknown	Epinephrine and thermocoagulation	Saraireh et al. [9]
BRBPR	74	Male	No	No	Arterial embolization	Ashour et al. [14]
Melena and hematochezia	81	Male	Unknown	Yes	Hemostatic clip	Dailey et al. [8]

NSAID, nonsteroidal anti-inflammatory drug.

Diagnosis of a Dieulafoy's lesion is usually performed via endoscopy [7]. Our patient was found to have a Dieulafoy's lesion within the cecum, an exceedingly uncommon and potentially underdiagnosed phenomenon. To our knowledge, there have been eight prior reported cases of GI bleeding secondary to a cecal Dieulafoy's lesion (Table 1). Disease presentation is variable with 62.5% presenting with painless hematochezia, 25% with BRBPR, and 12.5% with a mixed picture of melena and hematochezia [8–15]. Age at presentation ranged from 25 to 82 years. 62.5% of patients were males, further highlighting the approximate 2:1 ratio seen in Dieulafoy's lesions [8, 10, 11, 14, 15].

Treatment of Dieulafoy's lesions is contingent on the patient's clinical presentation, lesion location, and accessible expertise. Historically, Dieulafoy's bleeding was managed surgically [4]. Advances in endoscopic techniques over the years have shifted the management of Dieulafoy's-associated GI bleeding from surgery to endoscopy with its various techniques [16]. Such endoscopic techniques include epinephrine or ethanol injection, argon plasma coagulation, heater probe coagulation, hemostatic clips, or band ligation [7]. Three of the previously reported cases pertaining to cecal Dieulafoy's were successfully treated mechanically with hemostatic clipping, 2 with local epinephrine injection and thermocoagulation, 1 with right hemicolectomy, 1 with combination hemostatic clipping and ethanol injection, and 1 with arterial embolization [8–15]. When endoscopy is unsuccessful, angiography can be utilized in both Dieulafoy's lesion bleeding site localization and embolization, albeit with a risk of ischemia [17]. Due to ischemic risk, angiography is usually reserved for lesions unreachable by endoscopy and for patients who fail endoscopic therapy or are at an exceedingly high surgical risk [18, 19].

The etiology of Dieulafoy's lesions is not entirely understood. Proposed theories include mechanical pressure on the intestinal mucosa by the throbbing submucosal artery or arterial thrombosis, triggering necrosis of the arterial wall and eventually leading to rupture [3, 20]. Within lower GI tract Dieulafoy's lesions, irritation of the mucosa by transiting feces may also play a role [3]. There has been an association of antiplatelet, nonsteroidal anti-inflammatory drug, and alcohol use with Dieulafoy's lesions, though a causal relationship is

yet to be established [5, 21]. Aspirin use has also been associated with an increased risk of lower GI bleeding, albeit a relatively lower risk than that associated with upper GI hemorrhage [22, 23].

Our patient only experienced hematochezia days after dyspnea and fatigue onset. This unique course emphasizes the uncommon and potentially elusive presentation of Dieulafoy's lesions within the cecum. Dieulafoy's lesions of the lower GI tract should be considered when no clear culprit is discovered, particularly in the setting of concomitant use of antiplatelet agents and anticoagulants.

Statement of Ethics

Ethics approval was not required for this study in accordance with the local/national guidelines. Written informed consent was obtained from the patient for publication of the details of their medical case and any accompanying images.

Conflict of Interest Statement

All authors have no conflicts of interest to declare.

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No funding was received for this study.

Author Contributions

Amr Dokmak performed the literature search, interpreted the data, and drafted the manuscript. Ergen Muso revised the manuscript for critically important intellectual content and gave final approval of submitted version.

Data Availability Statement

All data generated or analyzed during this study are included in this article. Further inquiries can be directed to the corresponding author.

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