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Cecal Dieulafoy lesion is a rare cause of lower gastrointestinal bleeding: A case report

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Abstract

Dieulafoy lesions are a rare but life-threatening cause of gastrointestinal bleeding. Colonic Dieulafoy lesions are exceptionally rare, comprising only 2% of these lesions. We present a case of cecal Dieulafoy lesion as an unusual cause of lower gastrointestinal bleeding—along with hemoptysis. An 81-year-old male with pulmonary hypertension presented with a one-day history of hematochezia. He subsequently developed new small-volume hemoptysis/hematemesis with increasing oxygen requirements. Bronchoscopy revealed old blood in the left lower lobe, with no active bleeding. The hemoptysis was attributed to severe pulmonary hypertension. Colonoscopy revealed a 2-mm cecal Dieulafoy lesion with spurting bleeding, which was clipped. We report a rare case of cecal Dieulafoy lesion with only 13 other published cases. Our case was complicated by hemoptysis creating an interesting diagnostic dilemma. In patients bleeding from both oral and anal orifices, a brisk upper gastrointestinal bleed—as well as independent causes involving the gastrointestinal and respiratory tracts—should be considered.

Keywords

Lower gastrointestinal bleed, angiodysplasia, Dieulafoy lesion, hemoptysis, cecum

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Introduction

Acute gastrointestinal (GI) bleeding is a common cause of hospital admissions in the United States with an overall mortality rate of 5%–10%, which can vary depending on the etiology of the bleed.[1,2] Specifically, GI bleeds can be categorized as an upper GI bleed (UGIB) or a lower GI bleed (LGIB) depending on whether the origin of the bleed is proximal or distal of the ligament of Treitz, respectively. LGIBs account for 20%–30% of GI bleeds and occur more frequently in the elderly.[3] Indeed, LGIB has a greater than 200-fold increase in incidence from third to the ninth decade of life where age is also positively correlated with a longer hospital course.[2]

Diverticular disease and angiodysplasia are the predominant causes of LGIB in the elderly population.[2] Other common causes of LGIBs include ischemic colitis, inflammatory bowel disease, and malignancy.[3] Dieulafoy lesion, a specific type of angiodysplasia, is an uncommon but difficult to diagnose cause of severe, life-threatening UGIB or rarely LGIB.[4] Dieulafoy lesions, which are typically located in the stomach, esophagus, and duodenum and seldomly located in the colon, predispose for hemodynamically severe hemorrhage as these vessels maintain a constant arterial diameter despite their submucosal location, unlike other components of the arterial tree.[4] While UGIBs can be brisk, LGIBs may also present with massive bleeding in the elderly, especially in those with numerous comorbidities, where the mortality rate may be high as 21%.[3] Taken together, acute GI bleeds, encompassing both UGIB and LGIB, require prompt accurate diagnosis of the etiology and subsequent management to mitigate morbidity and mortality. Here, we present a case of a Dieulafoy lesion, located in the cecum, as a rare cause of LGIB.

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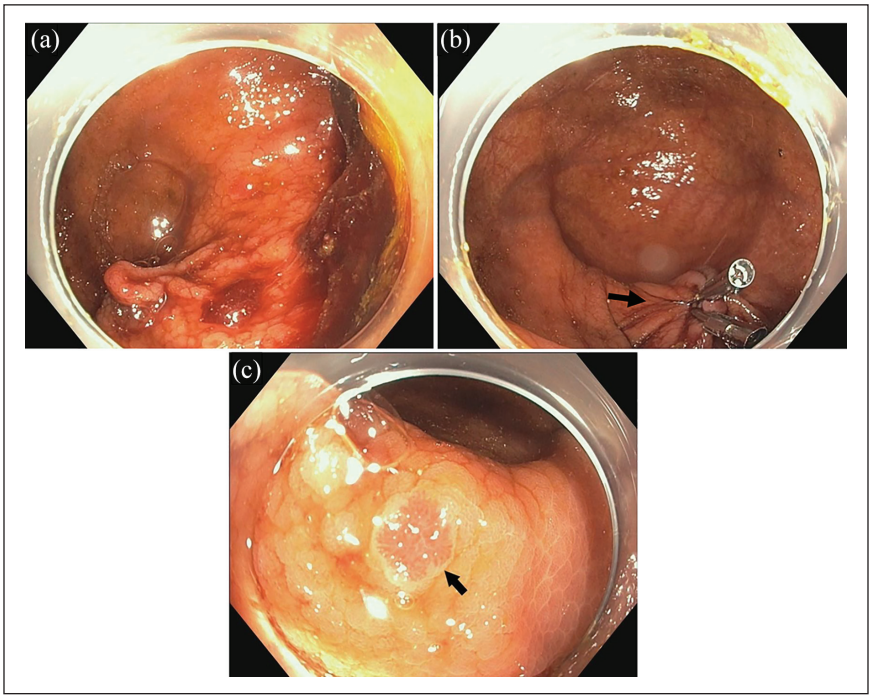

Figure 1. Colonoscopy findings in the cecum of a patient presenting with a lower gastrointestinal bleed. (a) Cecal Dieulafoy lesion. (b) Cecal Dieulafoy lesion after clipping (arrow). (c) Arteriovenous malformation (arrow).

Case

An 81-year-old male with a history of atrial fibrillation on warfarin, heart failure with moderately reduced ejection fraction (HFmrEF; EF 44%), coronary artery disease status post coronary artery bypass graft (CABG), pulmonary hypertension (pHTN; right ventricular systolic pressure [RVSP] 59 mmHg), moderate–severe tricuspid regurgitation (TR), obstructive sleep apnea, hypertension, hyperlipidemia, and thoracic aortic aneurysm presented with a one-day history of numerous grossly bright red bloody bowel movements. He also reported epigastric abdominal pain which had resolved, along with stable chronic dyspnea and leg swelling. He denied dyschezia, hematemesis, and lightheadedness. His last colonoscopy was performed 16 years prior to presentation.

On examination, he was afebrile (36.8°C), normotensive (101/63 mmHg), with normal heart (73 bpm) and respiratory rates (18 breaths/min) and oxygen saturation of 90% on home 3-L oxygen. Cardiopulmonary and abdominal examinations were normal. Digital rectal examination showed bright red blood with positive stool guaiac test but no external hemorrhoids. He had 3+ pitting edema of the lower extremities.

Laboratory studies revealed supratherapeutic international normalized ratio of 3.1, creatinine of 1.37 (baseline 1.1–1.3 mg/dL), and normal hemoglobin (12.9 g/dL). CT abdomen and pelvis was negative for evidence of GI bleeding.

Intravenous vitamin K was administered for warfarin reversal; however, he continued to have hematochezia. On day 2 of admission, he developed new small-volume hemoptysis/hematemesis with hemodynamic stability but had increasing oxygen requirements of 4–6 L. Chest imaging revealed a new, small, focal consolidative opacity in the left lower lung lobe representing possible hemorrhage. Bronchoscopy revealed old blood in the left lower lobe, with no active source of bleeding identified. On hospital day 3, colonoscopy revealed a solitary, 2-mm cecal Dieulafoy lesion with spurting bleeding which was clipped (Figure 1). Colonoscopy also revealed a nonbleeding deep mucosal arteriovenous malformation (AVM). Esophagogastroduodenoscopy showed erosive gastropathy in the gastric body and antrum. He had no recurrence of hematochezia nor hemoptysis and did not require any blood transfusions during his admission.

His hospital course was complicated by acute kidney injury, with peak creatine at 2.38 mg/dL. The cause was thought to be cardiorenal syndrome due to acute exacerbation of HFmrEF, given severe pitting edema on examination and new transthoracic echocardiogram showing enlarged inferior vena cava with reduced inspiratory collapse. Additionally, his RVSP had increased to 72 mmHg and the TR was now severe. He responded to diuresis and returned to home oxygen requirements, with an 8 kg reduction in weight upon discharge. He was transitioned to apixaban prior to leaving the hospital. On outpatient follow-up, he denied bleeding recurrence.

Discussion

The overall mortality from Dieulafoy lesion has decreased to 9%–13% from 30% a few decades ago, due to improved and

Table 1. Summary of cases of patients with cecal Dieulafoy lesion.

Case #	Author[Ref.]	Year of publication	Age (Years)	Sex (M/F)	Comorbidities	On anticoagulation (Y/N)	Hematemesis (Y/N)	Hemoptysis (Y/N)	Hematochezia or bleeding from the rectum (Y/N)	Melena (Y/N)	Hemodynamically stable (Y/N)	Required RBC transfusion (Y/N)	# of Units	Dx Tool	Overall Tx	Bleeding Recurrence (Y/N)	Prognosis: Survived Acute Event (Y/N)
1	Ashour[6]	2000	74	M	DM2	N	N	N	Y	N	N	Y	3	Angiography	Transcatheter Arterial Embolization	N	Y
2	Dailey[7]	2020	81	M	HTN, Hx PE	Y	N	N	Y	Y	NS	Y	1	Colonoscopy	Endoclips	N	Y
3	Farrell[8]	1992	77	M	Hx of PUD, Polycythemia Vera	NS	N	N	Y	N	N	Y	8	Angiography, Pathology	Right hemicolectomy	NS	Y
4			64	M	Alcoholic cirrhosis with varices, diffuse non-Hodgkin's Lymphoma	NS	N	N	N	Y	NS	Y	NS	Pathology post-mortem	NS	NA	N
5			80	F	Rheumatoid arthritis	NS	N	N	Y	N	N	Y	NS	Angiography, Pathology	Right hemicolectomy	NS	Y
6	Fuentes-Valenzuela[9]	2022	71	M	Pulmonary adenocarcinoma	NS	N	N	Y	N	N	NS	NA	Colonoscopy	APC + Endoclips	N	Y
7	Kinoshita [10]	2020	82	F	HTN, Hx CVA	N	N	N	Y	N	Y	Y	NS	Colonoscopy	Endoclips	N	Y
8	Pishori[11]	2003	25	M	NS	NS	N	N	Y	N	N	Y	4	Angiography, Pathology	Right hemicolectomy	N	Y
9	Ribeiro[12]	2021	85	F	HTN, HLD, CHF, CKD, Afib, Biologic Aortic Valve Prothesis	Y	N	N	Y	N	N	Y	5	Colonoscopy	Epi + Endoclips	N	Y
10	Saraireh[13]	2017	64	F	Hx colorectal adenocarcinoma	NS	N	N	Y	N	NS	NS	NA	Colonoscopy	Epi + Cauterization	N	Y
11	Singh[14]	2001	62	F	NS	NS	N	N	Y	N	N	Y	NS	Colonoscopy	Epi + Thermocoagulation	N	Y
12	Sone[15]	2000	34	M	NS	NS	N	N	Y	N	NS	Y	10	Colonoscopy	Epi + Hypertonic Saline + Ethanol + Endoclips	Y	Y
13	Tun[16]	2016	70	M	DM2, HTN	NS	N	N	Y	N	N	Y	8	Colonoscopy	Epi + Coagulation	N	Y
14	Our case	NA	81	M	Afib, CHF, CAD s/p CABG, pHTN, severe TR, OSA, HTN, HLD, thoracic aortic aneurysm	Y	N	Y	Y	N	Y	N	NA	Colonoscopy	Endoclips	N	Y

Afib: Atrial fibrillation; APC: argon plasma coagulation; CABG: Coronary artery bypass graft; CAD: Coronary artery disease; CHF: Congestive heart failure; CKD: Chronic kidney disease; CVA: Cerebrovascular accident; DM2: Diabetes Mellitus Type 2; Dx: Diagnostic; Epi: Epinephrine; HLD: Hyperlipidemia; HTN: Hypertension; Hx: History; NA: Not applicable; NS: Not stated; OSA: obstructive sleep apnea; PE: Pulmonary Embolus; pHTN: pulmonary hypertension; PUD: Peptic ulcer disease; RBC: red blood cell; TR: tricuspid regurgitation; Tx: Treatment..

aggressive endoscopic management.[4] However, these statistics are still concerning and warrant further recognition to decrease morbidity, mortality, and length of hospitalization.[4] Bleeding from Dieulafoy lesion is more prevalent among the elderly, possibly due to age-related mucosal wear and tear of the lesion.[4] Although Dieulafoy lesion itself is rare, it is commonly associated with and is responsible for 1.5% of UGIBs, given that 70% originate in the stomach with additional common sites including other upper GI tract locations.[4] While colonic lesions comprise only 2% of Dieulafoy lesions, it is important to include this rare phenomenon in the differential diagnosis for LGIB, to allow prompt management.[5] Here, we report a rare case of cecal Dieulafoy lesion as a cause for LGIB.

An extensive literature search for case reports in the English language of GI bleeding from Dieulafoy lesions in the cecum was conducted in PubMed/Medline and Embase since inception. This search yielded only 11 relevant articles consisting of 13 cases.[6–16] Only 3/13 cases were reported prior to 2000, which may signify increased awareness of Dieulafoy lesions as a rare cause of GI bleed (Table 1). Our case was similar to all 13 cases, which showed an average age of 67.9 ± 17.9 years and a male predominance (64.3%).[4] Additionally, women with Dieulafoy lesions were older and had smaller age variability (74.6 ± 10.8 years old) compared to men (64.1 ± 20.5 years old). This was due to two male outliers who were much younger than all other patients (Table 1).

As expected, most patients (11/14) presented with hematochezia/bleeding from the rectum only. However, one patient presented with melena and another patient, surprisingly, with both melena and hematochezia. Remarkably, ours was the only patient with both hemoptysis and hematochezia. Only two other patients, like ours, were on anticoagulation for indications including recent history of pulmonary embolism and atrial fibrillation. While in most of the cases the patients were hemodynamically unstable (8/14), only our patient and one other were hemodynamically stable throughout the hospitalization. Similarly, while our patient did not require a transfusion of packed red blood cells, all other cases that provided this information did require transfusions, where the average number of units required was 5.6.

Regarding diagnosis, Dieulafoy lesions were discovered mainly via colonoscopy (9/14); other modalities utilized included angiography +/− pathology (4/14). In one case, the Dieulafoy lesion was diagnosed postmortem. Most cases (9/14), including ours, were treated endoscopically with a combination of clips, epinephrine injections, and/or cauterization/coagulation. Less common treatment strategies included surgical management mainly via right hemicolectomy (3/14), followed by arterial embolization. In nearly all cases (10/14), including ours, management prevented rebleeding, and only one patient expired due to the acute GI bleed secondary to Dieulafoy lesion.

One limitation of our case was inability to ascertain the etiology of the hemoptysis, as the bronchoscopy revealed old blood without a distinct source of bleeding. However, the suspected cause of hemoptysis was significant pHTN or pulmonary hemosiderosis secondary to acute heart failure exacerbation, with preference for the former etiology given the severity of bleeding. Initially, it was not known whether the oral bleeding was due to hemoptysis or hematemesis, though hematemesis was initially considered given the presence of LGIB. This created an interesting but challenging diagnostic dilemma. We maintained a broad differential including a brisk UGIB with hematemesis and hematochezia, or two separate conditions causing hemoptysis and hematochezia. Unfortunately, this aspect remained unclear since the upper endoscopy showed erosive gastropathy and bronchoscopy old blood in the left lower lobe. Additionally, our patient was predisposed to bleeding from supratherapeutic anticoagulation and the presence of an AVM. While an AVM was discovered on colonoscopy, the bronchoscopy and chest imaging did not reveal similar findings in the pulmonary vasculature. Note that this paper was formatted according to the CARE guidelines for case reports.[17]

Conclusion

In conclusion, we present a rare instance of cecal Dieulafoy lesion and summarize 13 previous cases reported in the literature. Dieulafoy lesions are rare, and cecal Dieulafoy lesions are exceptionally rare. Our case was unique as it was complicated by subsequent hemoptysis/hematemesis, and while workup was suggestive of hemoptysis, this initially created an interesting diagnostic dilemma. Clinicians should recognize cecal Dieulafoy lesion as an unusual cause of LGIB. In patients with both oral and anal bleeding, a brisk UGIB should be considered. Depending on comorbidities, such are our patient with pHTN, both GI and respiratory sources of bleeding should be in the differential diagnosis.

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Author contributions

All authors contributed to the conceptualization and writing – review & editing. B.S. contributed to methodology, formal analysis and investigation, writing – original draft preparation; D.T. contributed to writing – original draft preparation; T.J.B. contributed to methodology and supervision.

Declaration of conflicting interests

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Ethics approval

Our institution does not require ethical approval for reporting individual cases.

Informed consent

Written informed consent was obtained from the patient for their de-identified information to be published.

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