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A rare cause of recurrent lower gastrointestinal hemorrhage due to a rectosigmoid Dieulafoy's lesion

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Abstract

Background Colonic Dieulafoy's lesions (DLs) are an extremely rare cause of acute gastrointestinal bleeding. However, it could be presented from self-limiting, mild bleeding episodes up to recurrent, massive life-threatening hemorrhage.

Case presentation We present a case of 9-year-old female with recurrent attacks of hematochezia. The colonoscopy showed actively bleeding Dieulafoy's lesions at the rectosigmoid colon that was successfully managed endoscopically by the application of two hemoclips.

Conclusion A prompt and accurate diagnosis in patients with a Dieulafoy's lesion carries paramount importance. A meticulous examination and proper resuscitation are a must because massive, uncontrolled bleeding from this lesion may culminate in the death of the patient fairly quickly.

Keywords Hematochezia, Dieulafoy's lesions, Hemoclips

Introduction

Dieulafoy's lesions (DL) also called caliber-persistent artery, submucosal arterial malformation, or solitary exulceration simplex are defined by a dilated aberrant submucosal vessel that erodes the overlying epithelium without obvious ulceration [1, 2]. On histological examination, there is no obvious abnormality in the arterial wall, and there is no evidence of vasculitis or arteriovenous shunting [3]. The exact pathophysiology of a Dieulafoy's lesion is still unknown [2]. Dieulafoy's lesions represent a rare cause for acute gastrointestinal bleeding that accounts for < 2% [4, 5]. DL are common in the proximal stomach along the lesser curvature near the esophagogastric junction, while they are extremely rare in the colon.

Endoscopy remains the first diagnostic modality; meanwhile, contrast-enhanced CT, angiography, and surgical interventions are preserved for patients with difficult-to-diagnose or in patients with a failed endoscopic diagnosis of management [2].

The difficulty in the endoscopic diagnosis of colonic DL is owing to the small size of the lesion and the insufficient visual field, especially in an acute setting, and poor preparation [6].

Here, we present the case of a 9-year-old female patient who experienced recurrent attacks of hematochezia secondary to colonic DL that was successfully managed endoscopically. This case highlights the importance of both careful endoscopic evaluation in investigating gastrointestinal hemorrhage and consideration of rare causes in the differential diagnosis of lower gastrointestinal (GI) bleeding.

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Case presentation

A female patient, 9 years old, from Kazakhstan presented to the emergency room (ER) with an attack of hematochezia. She reported at least two similar attacks over the last year for which she never sought medical advice. There was no associated nausea, vomiting, diarrhea, or abdominal pain. Her past medical and surgical history was unremarkable. The patient was admitted to the hospital for evaluation of her condition. She was vitally stable (BP was 110/70, PR 90 bpm) with unremarkable clinical examination. There was no history of bleeding from the skin or other orifices. Her laboratory investigations revealed a hemoglobin level of 13.5 g/dL (normal, 13.5–17.5 g/dL); platelet count of 230×10^9 (normal, $150–450\times10^9$/L); hematocrit 45% ($n=41–50\%$); international normalized ratio (INR) 1.02 ($n=0.8–1.1$); total leucocytic count 6.7×10^9 (normal, $3.5–10.5\times10^9$/L); C-reactive protein 5 ($n=$less than 10); blood urea nitrogen 20 mg/dL ($n=6–24$ mg/dL); creatinine 0.9 g/dL (normal, 0.6–1.2 mg/dL); serum sodium level 139 µ/L ($n=135–145$ µ/L); and serum potassium level 4.1 µ/L ($n=3.6–5.2$ µ/L).

An urgent colonoscopy was done, but unfortunately, the patient was very poorly prepared so she was planned for an elective colonoscopy after proper resuscitation and preparation. The second colonoscopy was done and revealed an active bleeding vascular lesion in the rectosigmoid colon (Figs. 1 and 2). Careful observation using the water immersion technique revealed pulsatile bleeding from a diminutive vessel surrounded by normal mucosa, which confirmed the diagnosis of Dieulafoy's lesion. No diverticula, hemorrhoids, angioectasias, or other mucosal lesions were identified. We successfully achieved endoscopic hemostasis with clipping (application of two hemoclips) (Fig. 3).

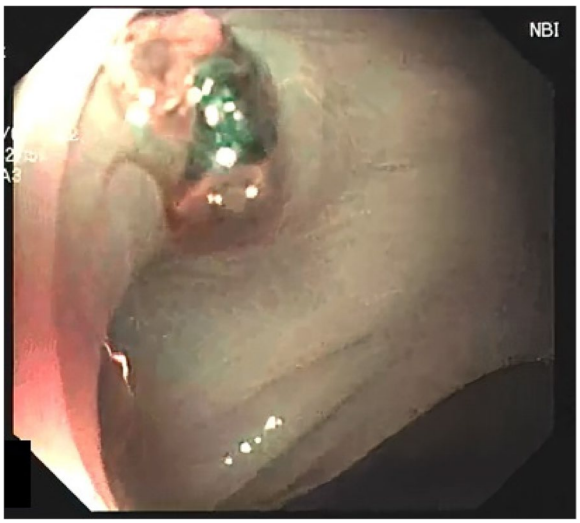

Fig. 2 Colonic Dieulafoy's lesion with narrowband image (NBI)

The patient did not experience any immediate or delayed post endoscopic complications, and she had no clinical or laboratory evidence of ongoing or recurrent bleeding.

Discussion

Colonic Dieulafoy's lesions (DL) are rare (all extragastric lesions ~ 5%), all are case reports, and they are one of the most common causes of hidden and recurrent bleeding. DLs "ulcers" account for less than 1% of GI hemorrhage.

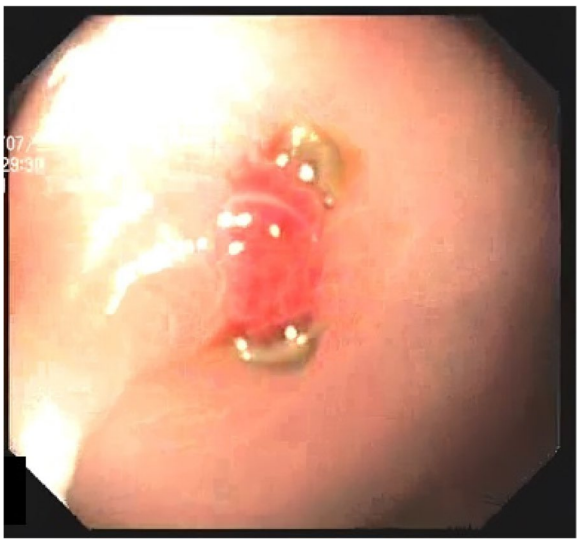

Fig. 1 The presence of active oozing blood from colonic Dieulafoy's lesion

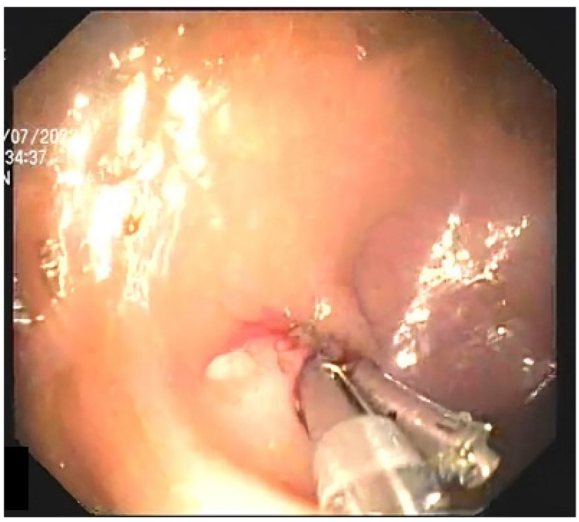

Fig. 3 Application of hemostatic endoclip to colonic Dieulafoy's lesion achieving hemostasis

Hematochezia accounts for only 6% of the presentations [1]. There was no age or gender predominance for DL; however, it is mentioned in some review articles that the mean age of presentation was 66 ± 17 years, with slight male predominance [7].

The diagnosis of a Dieulafoy's lesion poses a real significant challenge. The difficulty in diagnosis of DL comes from its common presentation with painless large-volume bleeding, which is difficult to distinguish from other causes of lower gastrointestinal bleeding such as arteriovenous malformations or diverticular hemorrhage. Even though the initial endoscopy may be efficient for the initial diagnosis of up to 70% of the patients, several endoscopies may be required if the source could not be identified. As reported in the literatures, about 6% of these patients may require three or more endoscopies to establish a diagnosis of DL; this failure may be explained by excessive blooding, blood clots, and/or subtle lesions [8]. The successful location of Dieulafoy's lesion could be also achieved with capsule endoscopy but the drawbacks of not allowing intervention [5].

Although there are no standard guidelines, the management of colonic Dieulafoy's lesions is predominantly endoscopic. Hemoclipping was always the best therapeutic modality, as other therapeutic modalities, including thermocoagulation, argon plasma coagulation, band ligation, and endoscopic sclerotherapy. However, the combination of multiple endoscopic techniques to achieve hemostasis, including clipping, adrenaline injection, and laser coagulation, has also been reported in the literature [9–11].

Rare cases were treated surgically with partial colectomy in the setting of uncontrollable, life-threatening lower gastrointestinal hemorrhage [9]. Selective angiography was also reported in patients who failed endoscopic therapy, for lesions beyond the reach of therapeutic endoscopy, or in patient's poor candidates for surgery [9].

Conclusion

This case illustrates a less common and more difficult-to-identify source of gastrointestinal bleeding, a Dieulafoy's lesion. Although rare, colonic Dieulafoy's lesions can be presented with life-threatening bleeding that should be considered in the differential diagnosis of acute lower gastrointestinal bleeding. Bad bowel preparation may mask diagnosis and further management. A good colonoscopy is a meticulous task.

Abbreviations

DL	Dieulafoy's lesions
GI	Gastrointestinal
ER	Emergency room
INR	International normalized ratio

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Authors' contributions

MN is the main endoscopist and read and revised the manuscript. AL is the assistant endoscopist and collected the data. AA wrote the manuscript and submission procedure. All authors read and approved the final manuscript.

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Availability of data and materials

Not applicable.

Declarations

Ethics approval and consent to participate

The study was approved by institution ethical committee and from review board of Kasr Al Ainy Hospital. Oral and written informed consents were obtained from the patient or from his eligible relatives.

Consent for publication

Oral and written informed consents were obtained from the patient or from his eligible relatives.

Competing interests

The authors declare that they have no competing interests.

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References

1. Nojkov B, Cappell MS (2015) Gastrointestinal bleeding from Dieulafoy's lesion: clinical presentation, endoscopic findings, and endoscopic therapy. World J Gastrointest Endosc 7(4):295–307. https://doi.org/10.4253/wjge.v7.i4.295
2. Inayat F, Ullah W, Hussain Q, Abdullah HMA. Dieulafoy's lesion of the colon and rectum: a case series and literature review. BMJ Case Rep. 2017;2017:bcr2017220431. Published 2017 Oct 25. https://doi.org/10.1136/bcr-2017-220431
3. Jain R, Chetty R (2009) Dieulafoy disease of the colon. Arch Pathol Lab Med 133(11):1865–1867. https://doi.org/10.5858/133.11.1865
4. Chaer RA, Helton WS (2003) Dieulafoy's disease. J Am Coll Surg 196(2):290–296
5. Baxter M, Aly EH (2010) Dieulafoy's lesion: current trends in diagnosis and management. Ann R Coll Surg Engl 92(7):548–554. https://doi.org/10.1308/003588410X12699663905311
6. Sudo G, Takagi H, Nakase H (2021) Lower gastrointestinal bleeding from Dieulafoy's lesion in the transverse colon. Dig Endosc 33(4):e56–e57. https://doi.org/10.1111/den.13936
7. Inayat F, Hussain A, Yahya S, et al. Rectal Dieulafoy's lesion: a comprehensive review of patient characteristics, presentation patterns, diagnosis, management, and clinical outcomes. Transl Gastroenterol Hepatol. 2022;7:10. Published 2022 Jan 25. https://doi.org/10.21037/tgh.2020.02.17
8. Inayat F, Amjad W, Hussain Q, Hurairah A. Dieulafoy's lesion of the duodenum: a comparative review of 37 cases. BMJ Case Rep. 2018;2018:bcr2017223246. Published 2018 Feb 22. https://doi.org/10.1136/bcr-2017-223246
9. Ma C, Hundal R, Cheng EJ. Colonic Dieulafoy's lesion: a rare cause of lower gastrointestinal hemorrhage and review of endoscopic management. Case Rep Gastrointest Med. 2014;2014:436293. https://doi.org/10.1155/2014/436293

10. Then EO, Bijjam R, Ofosu A, Rawla P, Culliford A, Gaduputi V. Rectal Dieulafoy's lesion: a rare etiology of lower gastrointestinal hemorrhage. Case Rep Gastroenterol. 2019;13(1):73–77. Published 2019 Feb 14. https://doi.org/10.1159/000497139
11. Fukita Y. Treatment of a colonic Dieulafoy lesion with endoscopic hemoclipping. BMJ Case Rep. 2013;2013:bcr2013009734. Published 2013 Apr 22. https://doi.org/10.1136/bcr-2013-009734

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