Case Report

Treatment of ileal Dieulafoy's lesion by hemostatic clips under double-balloon enteroscopy: a case report

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Background: Small intestinal Dieulafoy's lesion (DL) is a rare cause of life-threatening gastrointestinal bleeding. Based on previous case reports, the diagnostic approaches for DL located in jejunum and ileum are different. In addition, there is no available consensus regarding the treatment of DL, and previous case reports suggest that surgery is the preferable choice for small intestinal DL compared to endoscopic treatment. Notably, our case report indicates that double-balloon enteroscopy (DBE) should be an effective diagnostic and therapeutic approach for small intestinal DL.

Case Description: A 66-year-old female was transferred to the Department of Gastroenterology due to hematochezia and abdominal distension and pain for more than 10 days. She had a history of diabetes, hypertension, coronary heart disease, atrial fibrillation, mitral insufficiency, and acute cerebral infarction. Conventional diagnostic approaches, including gastroduodenoscopy, colonoscopy, and even angiogram, did not show any definite source of bleeding, and then a capsule endoscopy was performed and suggested that the bleeding may be located in ileum. Finally, she was successfully treated by hemostatic clips under DBE via anal route. And there is no recurrence after endoscopic treatment was observed in our case during a 4-month follow-up.

Conclusions: Although small intestinal DL is rare and difficult to be detected by conventional approaches, DL still needs to be considered as a differential diagnosis for gastrointestinal bleeding. In addition, DBE should be considered as a preferred choice for the diagnosis and treatment of small intestinal DL due to lower invasiveness and cost as compared to surgery.

Keywords: Dieulafoy's lesion (DL); small intestine; double-balloon enteroscopy (DBE); endoscopic treatment; case report

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Introduction

Dieulafoy's lesion (DL) is a developmental vascular malformation of the gastrointestinal tract, which was firstly described by a French surgeon in 1898 (1). The diameter of the lesion's artery, which ranges 1–3 mm, is almost 10 times that of normal arteries at the muscularis mucosae level, which may increase the risk of massive bleeding (2). The most common site of DL is the stomach, accounting for nearly three quarters, and the rarest site is the small intestine, only accounting for 1% (3). Herein, we reported a case with DL in ileum, which was then successfully treated by hemostatic clips under double-balloon enteroscopy (DBE). We present the following case in accordance with the CARE reporting checklist (available at https://tgh.amegroups.com/article/view/10.21037/tgh-22-14/rc).

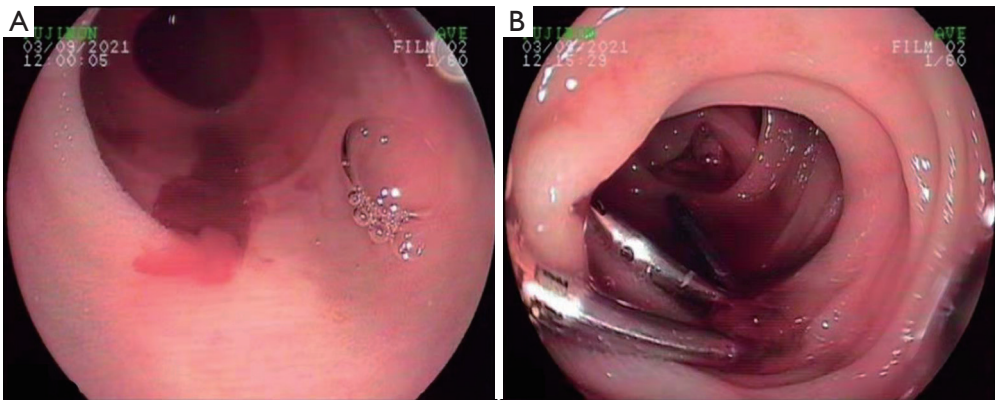

Figure 1 Diagnosis and treatment of DL in ileum under DBE. (A) DBE via anal route showed active pulsatile bleeding in the upper part of ileum; (B) three hemostatic clips were locally placed and 25,000 U thrombin was sprayed on this lesion to effectively treat this disease. DL, Dieulafoy's lesion; DBE, double-balloon enteroscopy.

Case presentation

On August 24, 2021, a 66-year-old female underwent endoscopy at outpatient department of our hospital due to hematochezia and abdominal distension and pain for more than 10 days. Gastroduodenoscopy did not find any sign of bleeding. Colonoscopy showed large blood clots in the intestinal tract, but did not detect any bleeding lesion. On August 28, 2021, she was admitted to the Department of General Surgery of our hospital. She had a history of diabetes, hypertension, coronary heart disease, atrial fibrillation, and mitral insufficiency, and a recent diagnosis of acute cerebral infarction for 10 days. And she had a medication history of aspirin, but aspirin had been stopped three months before this admission. Laboratory examinations showed hemoglobin (HGB) 71 g/L (reference range, 130–175 g/L), red blood cells (RBC) 2.44×10^{12}/L (reference range, $4.3–5.8\times10^{12}$/L), prothrombin time (PT) 15.1 s (reference range, 11.0–14.3 s), fibrinogen (FIB) 1.88 g/L (reference range, 2.00–4.00 g/L), antithrombin III (AT III) 69% (reference range, 80–120%), and D-dimer 1.54 μg/mL (reference range, 0.00–0.50 μg/mL). After pharmacological hemostasis, blood transfusion, and intravenous fluid supplementation, an angiogram was performed, but no source of bleeding was accurately identified.

On August 29, 2021, she was transferred to our department. After conservative therapy, HGB became up to 83 g/L. Considering that she had a recent history of acute cerebral infarction with coronary artery stenosis, a more invasive diagnostic approach, including DBE on general anesthesia, was compromised.

On August 31, 2021, the patient still presented with hematochezia. HGB was decreased to 66 g/L. Thus, a capsule endoscopy was performed and suggested that bleeding may be located in ileum. DBE via anal route was performed by an experienced endoscopist (F Gao), and showed active pulsatile bleeding in the upper part of ileum, without underlying ulcer, but with normal mucosa around the small defected mucosal lesions, which is consistent with a diagnosis of DL (*Figure 1A*). Then, 3 hemostatic clips were locally placed and 25,000 U thrombin was sprayed on the lesion to effectively treat this disease (*Figure 1B*). Finally, bleeding stopped.

The patient did not develop hematochezia after endoscopic treatment. On September 9, 2021, HGB was 94 g/L. The patient was discharged without any recurrence of gastrointestinal bleeding at a 4-month follow-up.

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

Discussion

DL, a rare cause of life-threatening gastrointestinal bleeding, constitutes about 1% to 2% of all episodes of gastrointestinal bleeding (1). In adults, the prevalence of DL in male versus female is 2:1 (4). In addition, DL can be found in all age groups, especially in the elderly with multiple

comorbidities, such as cardiovascular disease, hypertension, chronic kidney disease, and diabetes mellitus (5). Notably, multiple comorbidities were found in our patient.

Our patient presented with hematochezia. Initially, some possible causes for hematochezia were considered, mainly including diverticular bleeding, telangiectasias, vascular neoplasms, colitis, and malignancy. Finally, small bowel was considered as the potential location of bleeding in our patient. According to the clinical guidelines (6), patients with suspected small bowel bleeding should repeat gastroduodenoscopy or colonoscopy, if necessary. Meanwhile, hemodynamically stable patients with active bleeding can undergo computed tomography angiography (CTA) to identify the site of bleeding. Unfortunately, in our patient, conventional diagnostic approaches, including gastroduodenoscopy, colonoscopy, and even angiogram, did not show any definite bleeding source of gastrointestinal bleeding. Clinical guideline also recommends that video capsule endoscopy (VCE) should be firstly considered, if second-look gastroduodenoscopy and colonoscopy examinations are negative, and that computed tomographic enterography (CTE) or magnetic resonance (MR) should be performed in patients with suspected small bowel stenosis and negative capsule endoscopy (6). If CTE, MR, or VCE examinations are positive, deep enteroscopy should be further performed (6). In our patient, DBE via anal route was performed under the guidance of capsule endoscopy, and then showed that the location of bleeding should be ileum. Except for the approaches mentioned above, other advanced diagnostic approaches for detecting DL in small intestine were also employed in previous literature, including laparotomy, technetium-99m-labelled red blood cell scan (Tc^{99m}RBCs), and ileoscopy (*Table 1*) (7-31).

Until now, there is no available consensus regarding the treatment of DL. The treatment approaches for DL mainly include endoscopic and surgical treatments. Of the DL located in the upper gastrointestinal tract, 85% can be successfully treated by endoscopic therapy, and only 5% require surgery (32). However, it is difficult to detect DL located in the lower gastrointestinal tract, especially jejunum and ileum, by endoscopies, primarily due to residual stool and massive bleeding resulting in poor visualization of the gastrointestinal tract. Therefore, surgery is the preferred choice to treat small intestinal DL. Indeed, based on previous case reports (*Table 1*), surgery has been the most commonly used treatment approach for DL located in small intestine, but other approaches that are less invasive can be effective for the treatment of small intestinal DL. Approaches of endoscopic hemostasis mainly include mechanical hemostasis (i.e., hemostatic clips and endoscopic band ligation), regional injection (i.e., regional injection-epinephrine or norepinephrine injection and sclerotherapy), and argon plasma coagulation (APC) (1). In our patient, hemostatic clips were locally placed on the lesion to effectively treat ileal DL, which avoided surgery. In addition, angiography can be used to control bleeding by selective embolization of the feeding vessel (2).

Follow-up duration after successful treatment is heterogeneous among previous studies (*Table 1*), but no recurrence of DL is reported. Similarly, no recurrence after endoscopic treatment was observed in our case during a 4-month follow-up. Certainly, it is very necessary to monitor the risk of recurrent bleeding during the long-term follow-up in the future.

In conclusion, small intestinal DL is rare and difficult to be detected by conventional approaches. Notably, DBE is an effective diagnostic approach for DL in small intestine. Endoscopic treatment should be considered as a preferred choice of treatment with less invasiveness and lower cost as compared to surgery.

Table 1 Small intestinal DL: an overview of previous studies

First author (year)	Region	Number of patients	Sex (male/ female)	Age, years	Symptoms	Comorbidities	Approaches for detecting lesions	Lesion location	Treatment	Follow-up
Arai (2021)	Japan	1	M	66	Tarry stool	Cardiovascular disease and diabetes	Capsule endoscopy	Small intestine	APC	Alive 18 months
Wang (2021)	China	1	M	7	Melena	Hypertension	Enteroscopy	Jejunum	Surgery	Alive 12 months
Salimi (2021)	Iran	1	M	68	Hematemesis	No	CTA	Small intestine	Surgery	Alive 36 months
Saada (2019)	Israel	1	M	27	Bloody diarrhea	NA	Tc^{99m}RBCs and Laparotomy	Jejunum	Surgery	Alive 3 months
Kawabata (2019)	Japan	1	M	81	Tarry stool	Cerebral infarction	Enhanced CT and Enteroscopy	Jejunum	Hemoclips	Death unrelated to DL 10 weeks
Kieswetter (2019)	Canada	1	F	9	Melena and fresh blood	No	Angiography and enteroscopy	Jejunum	Surgery	Alive 48 months
Jung (2019)	Germany	1	NA	69	Melena	No	Enteroscopy	Jejunum	OTSC clip	Alive 6 days
Zhao (2018)	China	1	M	41	Hematochezia	NA	Enteroscopy	Jejunum	Surgery	Alive 6 months
Becq (2018)	France	1	F	32	Hematochezia	No	CTA and endoscopy	Ileum	Surgery	NA
Seo (2017)	Korea	1	M	25	Hematochezia	No	Angiography and capsule endoscopy	Jejunum	Surgery	Alive 24 months
Chen (2016)	China Taiwan	1	M	54	Bloody stool	Hypertension	Enteroscopy and CTA	Ileum	Hemoclips and surgery	Alive 3 months
Ego (2015)	Japan	1	F	95	Melena	Chronic cardiac failure and chronic kidney disease	Enteroscopy	Jejunum	Endoscopic band ligation	Alive 1 month
Shibutani (2011)	Japan	1	F	14	Hematochezia	No	CT scan and angiography	Ileum	Surgery	NA
Saji (2010)	Japan	1	M	72	Melena	Hypertension	Angiography, CT scan, and gastrointestinal endoscopy	Jejunum	Epinephrine injection and hemoclips	NA
Moreira-Pinto (2009)	Portugal	1	F	14	Hematochezia	NA	Laparotomy	Jejunum	Surgery	Alive 48 months

Table 1 *(continued)*

Table 1 *(continued)*

First author (year)	Region	Number of patients	Sex (male/ female)	Age, years	Symptoms	Comorbidities	Approaches for detecting lesions	Lesion location	Treatment	Follow-up
Yano (2008)	Japan	6	M	58	NA	NA	Enteroscopy	Jejunum	APC	NA
			M	68	NA	NA	Enteroscopy	Jejunum	APC and epinephrine injection	NA
			F	72	NA	NA	Enteroscopy	Jejunum	Hemoclips	NA
			M	56	NA	NA	Enteroscopy	Jejunum	Hemoclips	NA
			F	71	NA	NA	Enteroscopy	Ileum	Hemoclips	NA
			M	24	NA	NA	Enteroscopy	Jejunum	Surgery	NA
Tsai (2007)	China Taiwan	1	M	2	Dark-red stool	NA	Angiography	Ileum	Surgery	NA
Palma (2006)	Italy	1	M	50	Melena	Cardiovascular disease	Capsule endoscopy and enteroscopy	Small intestine	Hemoclips	Alive 6 months
Kim (2005)	Korea	1	M	35	Dizziness and palpitation	NA	CT scan	Jejunum	Surgery	NA
Mino (2004)	Japan	1	F	31	Melena	NA	Tc^{99m}RBCs and angiography	Jejunum	Surgery	Alive 36 months
Morowitz (2004)	America	1	M	4	Hematochezia	No	Laparotomy	Ileum	Surgery	NA
Iglesias (2004)	Spain	1	M	68	Rectal bleeding	NA	Ileoscopy	Ileum	Hemoclips and epinephrine injection	Alive 12 months
Fox (2001)	Britain	1	F	47	Melaena	Hypertension	Laparotomy	Ileum	Surgery	NA
Blecker (2001)	America	2	M	18	Maroon colored stool	No	Tc^{99m}RBCs and laparotomy	Jejunum	Surgery	NA
			F	69	Abdominal pain	Peptic ulcer disease	Enteroscopy and laparotomy	Ileum	Surgery	NA
Lee (1997)	Korea	1	F	20	Hematochezia	NA	Tc^{99m}RBCs	Jejunum	Surgery	Alive 2 months

DL, Dieulafoy's lesion; APC, argon plasma coagulation; OTSC, over-the-scope; CT, computed tomography; CTA, computed tomography angiography; Tc^{99m}RBCs, technetium-99m-labelled red blood cell scan; NA, not applicable.

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Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at https://tgh.amegroups.com/article/view/10.21037/tgh-22-14/rc

Peer Review File: Available at https://tgh.amegroups.com/article/view/10.21037/tgh-22-14/prf

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://tgh.amegroups.com/article/view/10.21037/tgh-22-14/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

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