CASE REPORT

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Ileal Dieulafoy Lesion in a Patient with Glanzmann Thrombasthenia Presented with Hematochezia

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ABSTRACT Dieulafoy lesion (DL) is an important cause of acute gastrointestinal hemorrhage (GIH) because endoscopic diagnosis is very difficult. We report an ileal DL in a patient with Glanzmann Thrombasthenia (GT). A forty-year old female presented with bloody stool. Records showed that she was diagnosed with GT at 3 years of age. Physical examination on admission, she was clinically stable, except for tachycardia (110 bpm). Rectal examination revealed bright red blood. Her haemoglobin level was 8.7 g/dl. Colonoscopy revealed a DL at the terminal ileum. It was treated with combined endoscopic therapy of epinephrine injection followed by two clips application. Haemoglobin values were stable in the post-procedural period. DL in the terminal ileum is very rare. There have been fewer than 20 cases reported in the literature to date. Also GT is a rare congenital bleeding disorder. This case report revealed the coincidence of two rare disorders.

Keywords: Gastrointestinal hemorrhage; dieulafoy; ileum; endoscopy; thrombasthenia

Dieulafoy lesion (DL) is a vascular malformation.[1] It was first described by Paul Georges Dieulafoy in 1989.[2] It is usually found in the stomach. But, it can also occur anywhere in the gastrointestinal (GI) tract, such as the small bowel.[3] Endoscopic diagnosis is very difficult especially on the first episode of acute gastrointestinal hemorrhage (GIH). Endoscopic treatments are very effective and successful in controlling bleeding. Especially combination endoscopic therapy is superior to monotherapy because of a lower rate of recurrent bleeding.[4] We report an ileal DL, which was successfully treated with combined dual endoscopic therapy.

CASE REPORT

A forty-year old female presented with bloody stool that had begun one day earlier. Records showed that she was diagnosed with Glanzmann Thrombasthenia (GT) at 3 years of age. She was using no drugs except oral contraceptives. Physical examination on admission, arterial blood pressure at admission was 128/72 mmHg, and heart rate was 110 bpm. The nasogastric aspirate was clear. Rectal examination revealed bright red blood. Her haemoglobin level was 8.7 g/dl, platelet count was 103 000/mm^3. The result was rechecked and revealed a platelet count of 180 000/mm^3. She received tranexamic acid and one unit of apheresis platelet. Colonoscopy

revealed a DL on the terminal ileum approximately 2 cm from the ileocecal valve (Figure 1). It was treated with dual endoscopic therapy of epinephrine injection (1:10.000 solution) followed by two clips application (Olympus HX-110UR) (Figure 2) (Video). Haemoglobin values were stable in the post-procedural period and patient had had no recurrence of bleeding at a 3-month follow-up visit. Informed consent was taken from the parents.

DISCUSSION

DL is a rare cause of acute GIH (1-2%).[1] These are most frequently observed in the upper GI tract, particularly in the lesser curvature.[4] However, they can be observed in the small intestines as well, being most commonly in jejunum and rarely in the ileum.[3] There have been fewer than 20 cases reported in the literature to date.[5-10] Despite its rarity, DL should always be considered in the differential diagnosis of lower GIH. Endoscopy has advanced in recent years and endoscopic treatment is very successful in terms of achieving hemostasis.[10] Combined endoscopic therapy consists of injection therapy followed by thermal or mechanical therapy proved significantly superior to epinephrine injection alone. Angiography may be used for both localization and also therapy. If the rate of bleeding is massive or after failure of therapeutic endoscopic and angiographic interventions, surgical intervention can be used.[11]

GT is a rare congenital bleeding disorder and GI tract is the site of bleeding in about 10% of cases. Most common causes of GIH are gastroduodenal chronic lesions (gastric and duodenal ulcers) and from acute lesions (erosive acute gastritis).[12] Here, we think this case revealed the coincidence of two rare disorders. Also we observed only a temporary control of the GIH in our patient due to the past medical history of bleeding diathesis. In this setting, mechanical therapy (clip or band ligator) is one of the best modalities in active bleeding with a visible vessel.[13]

In summary, we showed that ileal DL in a patient with GT presented with hematochezia. It was treated with dual endoscopic therapy.

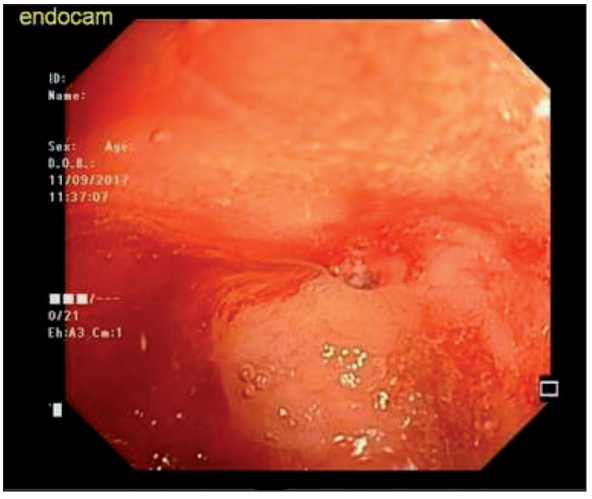

FIGURE 1: Colonoscopy revealed a Dieulafoy lesion on the terminal ileum.

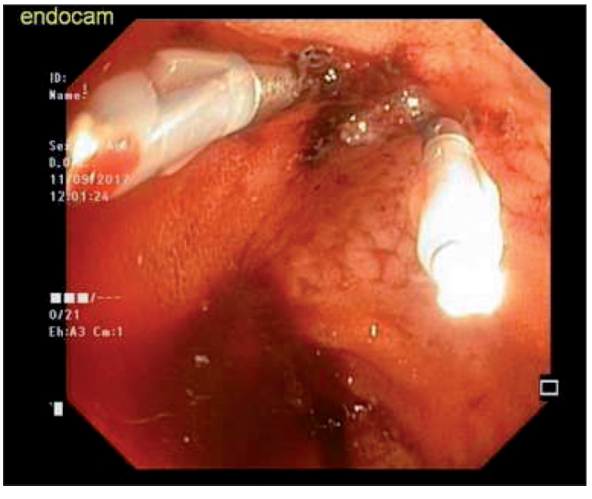

FIGURE 2: Dieulafoy lesion was treated with injection of epinephrine and followed by two clips application.

Source of Finance

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: *Abdullah Murat Buyruk, Ömer Özütemiz;* ***Design:*** *Abdullah Murat Buyruk, Nilay Danis;* ***Control/Supervision:*** *Abdullah Murat Buyruk, Ömer Özütemiz;* ***Data Collection and/or Processing:*** *Abdullah Murat Buyruk, Ömer Özütemiz;* ***Analysis and/or Interpretation:*** *Abdullah Murat Buyruk, Nilay Danis, Ömer Özütemiz;* ***Literature Review:*** *Abdullah Murat Buyruk;* ***Writing the Article:*** *Abdullah Murat Buyruk, Nilay Danis;* ***Critical Review:*** *Abdullah Murat Buyruk;* ***References and Fundings:*** *Abdullah Murat Buyruk;* ***Materials:*** *Abdullah Murat Buyruk.*

REFERENCES

1. Baxter M, Aly EH. Dieulafoy's lesion: current trends in diagnosis and management. Ann R Coll Surg Engl. 2010;92(7):548-54. [Crossref] [PubMed] [PMC]
2. Rodney E, Nihar S, Joseph RD. Gastrointestinal Bleeding Due to a Dieulafoy Lesion in the Afferent Limb of a Billroth II Reconstruction. Gastroenterol Hepatol (NY) 2011;7(4):268-71.
3. Dulic-Lakovic E, Dulic M, Hubner D, Fuchssteiner H, Pachofszky T, Stadler B, et al. Bleeding Dieulafoy lesions of the small bowel: a systematic study on the epidemiology and efficacy of enteroscopic treatment. Gastrointest Endosc. 2011;74(3):573-80. [Crossref] [PubMed]
4. Nojkov B, Cappell MS. Gastrointestinal bleeding from Dieulafoy's lesion: Clinical presentation, endoscopic findings, and endoscopic therapy. World J Gastrointest Endosc. 2015;7(4):295-307. [Crossref] [PubMed] [PMC]
5. Choi YC, Park SH, Bang BW, Kwon KS, Kim HG, Shin YW. Two cases of ileal dieulafoy lesion with massive hematochezia treated by single balloon enteroscopy. Clin Endosc. 2012;45(4):440-3. [Crossref] [PubMed] [PMC]
6. Fox A, Ravi K, Leeder PC, Britton BJ, Warren BF. Adult small bowel Dieulafoy lesion. Postgrad Med J. 2001;77(914):783-4. [Crossref] [PubMed] [PMC]
7. UEG Week 2015 Poster Presentations. United European Gastroenterol J. 2015;3(5 Suppl): 146-687. [Crossref] [PubMed]
8. Ezzat RF, Hussein HA, Baban TS, Rashid AT, Abdullah KM. Typhoid ulcer causing life-threatening bleeding from Dieulafoy's lesion of the ileum in a seven-year-old child: a case report. J Med Case Rep. 2010;4:171. [Crossref] [PubMed] [PMC]
9. Becq A, Dray X, Boarini G. Massive ileal bleeding secondary to a Dieulafoy's lesion. Endosc Int Open. 2018;6(4):E495-7. [Crossref] [PubMed] [PMC]
10. Jeon HK, Kim GH. Endoscopic management of Dieulafoy's lesion. Clin Endosc. 2015;48(2): 112-20. [Crossref] [PubMed] [PMC]
11. Lee YT, Walmsley RS, Leong RW, Sung JJ. Dieulafoy's lesion. Gastrointest Endosc. 2003;58(2):236-43. [Crossref] [PubMed]
12. Calabrese C, Di Febo G, Areni A, Morelli MC, Scialpi C, Brandi G, et al. Severe and relapsing upper gastrointestinal bleeding in a patient with Glanzmann's thrombasthenia. Dig Dis Sci. 2000;45(3):633-6. [Crossref] [PubMed]
13. Raju GS, Gajula L. Endoclips for GI endoscopy. Gastrointest Endosc. 2004;59(2): 267-79. [Crossref]