Archives of Academic Emergency Medicine. 2022; 10(1): e61

CASE REPORT

Jejunal Dieulafoy's Lesion as a Rare Cause of Massive Gastrointestinal Bleeding; a Case Report and Literature Review

Mohammad Ebrahim Kalantari[1], Newsha Sardarzadeh[1], Ali Mirsadeghi[2], Ali Akbar Bagherzadeh[3], Tooraj Zandbaf[2]*

1. Surgical Oncology Research Center, Mashhad University of Medical Sciences, Mashhad, Iran.
2. Department of General Surgery, Faculty of Medicine, Mashhad Medical Sciences, Islamic Azad University, Mashhad, Iran.
3. Innovative Medical Research Center, Faculty of Medicine, Mashhad Medical Sciences, Islamic Azad University, Mashhad, Iran.

Received: May 2022; Accepted: June 2022; Published online: 27 July 2022

Abstract: Jejunal Dieulafoy's lesion is difficult to diagnose due to its rarity, intermittent hemorrhage, and lesion site, which is largely inaccessible to conventional endoscopes. A 39-year-old man, who had no underlying disease, presented to the emergency department (ED) with weakness, dizziness, and dry cough with a history of several rectal bleeding episodes in the last few years. Endoscopy was normal, and the colon was full of clots on colonoscopy, and no gross pathology was found. On computed tomography (CT) angiography, a hyperdensity was seen in the middle of the jejunum, possibly suggesting contrast extravasation. Due to decreased hemoglobin of the patient, and hemodynamic instability, the patient became a candidate for surgery. A palpable lesion in the Jejunum was touched that opened longitudinally, which revealed active arterial bleeding from the nipple-like lesion. This segment was resected, and an anastomosis was performed. Histopathological examination of the small intestine confirmed a Dieulafoy's lesion.

It seems that, when upper endoscopy and colonoscopy fail to identify the cause of gastrointestinal bleeding, a Dieulafoy's lesion should be included in the differential diagnoses.

Keywords: Dieulafoy's lesion; Jejunal Diseases; Intestine, Small; Gastrointestinal Hemorrhage; Jejunum

Cite this article as: Kalantari ME, Sardarzadeh N, Mirsadeghi A, Bagherzadeh AA, Zandbaf T. Jejunal Dieulafoy's Lesion as a Rare Cause of Massive Gastrointestinal Bleeding; a Case Report and Literature Review. Arch Acad Emerg Med. 2022; 10(1): e61. https://doi.org/10.22037/aaem.v10i1.1623.

1. Introduction

Dieulafoy's lesion (DL) was first described as "miliary aneurysms of the stomach" by Dr. M. T. Gallard in 1884 (1). In 1898, the French surgeon Paul Georges Dieulafoy gave a more detailed description of this clinical condition, calling it an "exulceratio simplex"(2). In 1944, Levine and Valk were the first to describe a jejunal aneurysm with a submucosal artery rupture (3). The jejunum is where 1% of all confirmed cases of DL are identified (4). Jejunal DL is difficult to diagnose due to its rarity, intermittent hemorrhage, and lesion site, which is largely inaccessible to conventional endoscopes (5).

With a male-to-female ratio of 1.2:1, Jejunal DL has a little male predominance, with ages ranging from 10 to 95 years (55 ± 24 years; median, 82 years). The majority of individuals were diagnosed between the ages of 70 and 80. Most instances were found in developed countries. Several patients were on anticoagulants and/or nonsteroidal anti-inflammatory drugs due to their older age and underlying chronic medical issues (6).

DL is a rare and uncommon cause of massive gastrointestinal (GI) bleeding, and therefore, we would like to present our clinical experience with this unusual case to get more familiar with its various facets. Our work has been documented according to the CARE guidelines.

***Corresponding Author:** Tooraj Zandbaf; Faculty of Medicine, Bazarche Sarab, Imam Khomeini 14, Mashhad, Iran. tooraj.zandbaf@gmail.com, https://orcid.org/0000-0002-4882-5058, Tel:+98-9155114523; +98-513-2250041, Fax Number: +98-513-2250048, ORCID: https://orcid.org/0000-0003-2840-056X.

2. Case presentation

The patient was a 39-year-old man who presented to the emergency department (ED) with rectal bleeding, weakness, dizziness, and dry cough. The patient had a history of several rectal bleeding episodes in the last few years, as well as oral opium consumption. No history of underlying diseases was reported. An upper endoscopy had been performed a year ago, in which no lesions were seen. The patient was referred to our center from another hospital, where he had received 4 packed cells due to rectal bleeding.

An initial evaluation in the ED revealed a normal temperature of 37.3°C, blood pressure of 90/60 mmHg, heart rate of 110 beats per minute, respiratory rate of 28 breaths per minute, and normal oxygen saturation of 96% in room air. Lung examination was not remarkable. On abdominal examination, there was a slight generalized tenderness. The blood test results of the patient are presented in Table 1. We think the cause of the patient's thrombocytopenia was previous transfusion of packed cells. Sputum polymerase chain reaction (PCR) for SARS-CoV-2 infection was positive. The patient was admitted in COVID-19 ward and was resuscitated using intravenous crystalloids and four units of packed cells after a central venous (CV) line was inserted. A chest x-ray was taken from the patient, which indicated homogeneous opacity in the lower zone of both lungs (Figure 1A). The internist visited the patient and asked for a consultation with a general surgeon due to the lack of severe pulmonary problems as well as the significant rectal bleeding. Consultation was performed with a general surgeon, and computed tomography (CT) angiography and gastroenterologist consultation were requested.

Endoscopy was normal and on colonoscopy, the colon was full of clots, which made it impossible to examine the colon closely. At this time, CT angiography was performed. Hyperdensity was seen in the middle of the jejunum loops, possibly suggesting contrast extravasation. A small volume of free fluid was seen in the abdominal and pelvic cavities (Figure 1B). At this time, the patient began to decompensate, and the patient's hemoglobin decreased, resulting in need for additional packed cells and surgery. After laparotomy and during abdominal exploration, a palpable lesion in the small intestine was touched in 90 cm of ligament Treitz. At the site of the lesion, the small intestine opened longitudinally, which revealed active arterial bleeding from the nipple-like lesion. This segment of the small intestine was resected, and an anastomosis was performed. No other pathology was found in the abdomen.

The patient was transferred to the intensive care unit (ICU). The patient's gastrointestinal bleeding was stopped, and hemodynamics stabilized. After establishing bowel habits, the liquid diet was started, and finally, the patient was discharged in good general condition one week after surgery. Histopathological examination of the small intestine confirmed a DL.

Table 1: The laboratory findings of the patient

Blood Test	Result	Normal Range
White Blood Cells ($x10^3/\mu l$)	17.3	4-10
Hemoglobin (g/dl)	5.3	13.3-17.2
Hematocrit (%)	16	38.9-50.9
Platelets ($x10^3/\mu l$)	58	150-450
ESR1(mm/h)	3	<20
CRP2 (mg/L)	16.8	Up to 5.0
Blood Sugar(mg/dl)	112	75-100
Sodium(mEq/L)	130	136-146
Potassium(mEq/L)	6.3	3.5-5.5
D-Dimer (ng/FEU ml)	946	<500
Blood Urea Nitrogen(mg/dl)	59	10-50
Creatinine(mg/dl)	1.5	0.7-1.4
Alanine Aminotransferase (IU/L)	73	Up to 41
Aspartate Aminotransferase (IU/L)	88	Up to 37
Amylase (IU/L)	443	0.00-86.00
Prothrombin Time (Seconds)	17.3	10.5-14
Partial Thromboplastin Time (Seconds)	28	10-40
INR[3]	1.6	-

1: Erythrocyte Sedimentation Rate. 2: C-Reactive Protein.
3. International Normalized Ratio.

3. Discussion

Jejunal DL is an unusual cause of gastrointestinal bleeding, which can be severe and fatal. DL is a vascular anomaly with a diameter of 1 to 3 mm, which is 10 times bigger than the size of a healthy submucosal artery (7). A tiny mucosal defect with no surrounding mucosal inflammation characterizes this condition. These abnormal arteries usually take a tortuous path and protrude (2-5 mm) through the mucosal defect, making them prone to slight mechanical damage (8). Antiplatelet drugs and alcohol intake were found to be risk factors for DL formation in the upper gastrointestinal tract. The mean hemoglobin level in patients with jejunal DL was found to be 8.4 g/dL at the time of presentation. The quantity of blood units given out varied from two to nine. The possible presenting symptoms of dizziness/fatigue (11%) and iron-deficiency anemia can be explained by the decline in hemoglobin levels (10%) (6).

Jejunal DL remains difficult to diagnose. The intermittency of the bleeding and lack of access to the jejunum with a regular endoscope makes it difficult to detect using conventional endoscopy. Endoscopic guidance in the narrow jejunal canal can also be challenging (9). In our patient, we faced diagnostic challenge and a variety of differential diagnoses were considered when endoscopy and colonoscopy failed to identify the source of gastrointestinal bleeding.

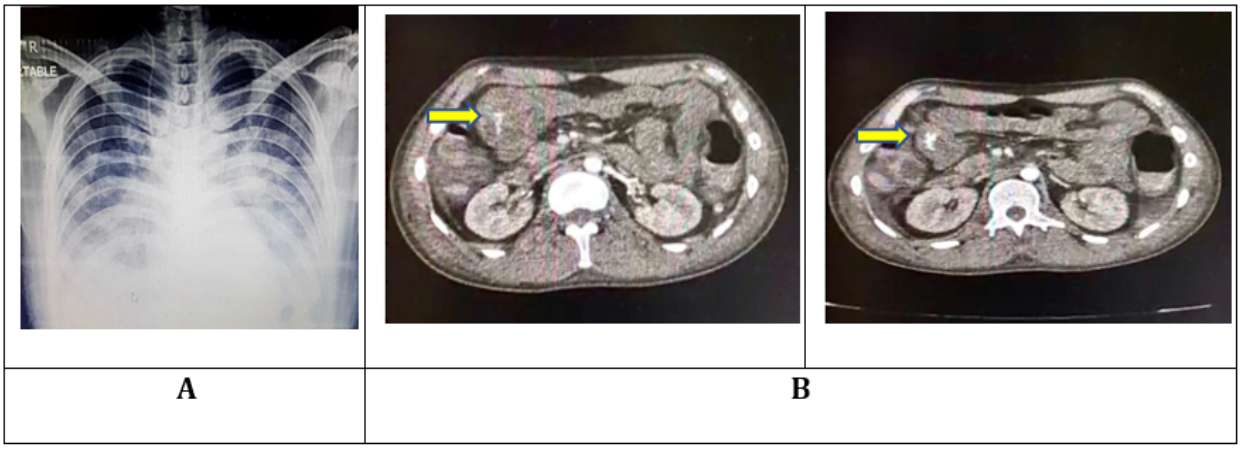

Table 2: Posterior-anterior Chest x-ray of the patient with homogeneous opacity in the lower zone of both lungs (A); Computed tomography angiography of patient suggesting contrast extravasation (B).

The diagnosis can be further complicated by the presence of ambiguous clinical symptoms that could be mistaken for peptic ulcer disease or gastrointestinal malignancies. As a result, the efficiency of traditional endoscopy in Jejunal DL is debatable (6). In our patient, after conventional endoscopies failed to detect the origin of bleeding, small bowel tumor was the most likely differential diagnosis we assumed.

Mesenteric angiography can be useful in cases of unidentified cause of bleeding when hemodynamic instability is evident. The use of computed tomography angiography to detect extra-gastric culprit vessels is very important. CT angiography is a quick and simple method that can identify bleeding rates as low as 0.5 mL/minute (10). The most sensitive tool for diagnosing active GI bleeding is radionuclide studies, which may detect bleeding rates as low as 0.1 mL/minute (11).

Capsule endoscopy and push enteroscopy have also been tried, with mixed results, particularly in stable patients who had unremarkable initial endoscopies. Single and double balloon enteroscopy are the best procedures with diagnostic yields of 96% and 98%, respectively. Since they complement noninvasive procedures like capsule endoscopy, these device-assisted enteroscopies are more effective in identifying DL. They provide secure and efficient diagnostic and therapeutic access to the jejunum via deep direct endoscopic access. The clinical presentation and capsule endoscopy results are important in determining oral or anal approach (6,12). Endoscopic therapy was initially used to treat 64% of patients. In 32% of cases, direct surgical therapy was used, while angiographic embolization was performed in 4% of cases. In most hemodynamically unstable individuals, surgical assessment is required. Intraoperative enteroscopy can be used to help locate the bleeder during surgery (6).

Yehya et al. presented a 19-year-old guy with multiple episodes of hematemesis and hematochezia. After resuscitation, a CT angiography was performed, which revealed no areas of active extravasation. The results of emergency upper and lower endoscopy were unclear. A tagged red blood cell scan revealed active bleeding in the proximal jejunum. The patient began to decompensate, necessitating additional blood products and vasopressors. Immediate operative intervention was accomplished and the bleeding Dieulafoy's lesion in the jejunum was discovered. Intestinal resection and primary anastomosis were performed (11).

Saada et al. presented a 27-year-old man with acute abdomen, bloody diarrhea, and syncope. Upper and lower endoscopies, as well as angiography, failed to reveal a bleeding location during the investigation. Following that, a scintigraphy revealed increased radiotracer activity in the right abdomen, which was consistent with small intestinal bleeding. Based on these findings, the patient underwent an emergency laparotomy, which revealed the bleeding location in the jejunum and allowed for surgical resection (13). Unlike these two cases, our patient was in his fourth decade of life and his CT angiography showed contrast extravasation in the jejunum. Similarly, our patient's gastrointestinal bleeding was surgically controlled.

An obscure gastrointestinal bleeding occurs when the cause of bleeding cannot be identified using upper endoscopy or colonoscopy, and a DL, among other conditions, should always be included in the differential diagnosis. Before any surgical intervention, if the patient's condition is stable and there is enough time, CT angiography or radionuclide scan probably help identify the source of the bleeding and assist the surgeon during the operation.

4. Conclusion

When upper endoscopy and colonoscopy fail to identify the cause of gastrointestinal bleeding, a DL should be included in the differential diagnosis. Surgery for a DL is now uncommon due to significant developments in endoscopic methods. If endoscopy fails to locate the source of bleeding, further imaging can be performed. In urgent circumstances, where additional imaging is not possible owing to a lack of time or resources, appropriate excision of the lesion for complete remission of the disease process should be undertaken.

5. Declarations

5.1. Acknowledgments

The authors would like to express their profound gratitude to medical personnel involved in the treatment of this patient in Imam Reza Hospital.

5.2. Informed consent

The patient's written consent was obtained for the publication of this case report.

5.3. Conflict of Interest

None.

5.4. Funding and supports

None.

5.5. Competing interests

The authors declare that they have no competing interests.

5.6. Authors' contributions

MEK and NS performed the surgery and reviewed the literature, TZ, AM, and AAB wrote the manuscript and reviewed the literature. All authors read and approved the final manuscript.

References

1. Gallard T. Aneurysmes milliares de l'estomac, donnant lieu a des hematemeses mortelles. Bull. Mem. Soc. Med. Hop. Paris. 1884; 1:84-91.
2. Dieulafoy G. Exulceratio simplex. L'intervebtion chirurgicale dans les hematemeses foudroyantes consecutives a l'exulceration simple de l'estomac. Bull de l'Acad de Med. 1898; 39:49-84.
3. Levine J, Valk AD. Aneurysm with rupture of a submucosal artery in the jejunum: case report. American Journal of Clinical Pathology. 1944 Nov 1;14(11):586-9.
4. Nojkov B, Cappell MS. Gastrointestinal bleeding from Dieulafoy's lesion: clinical presentation, endoscopic findings, and endoscopic therapy. World journal of gastrointestinal endoscopy. 2015 Apr 16;7(4):295.
5. Schmulewitz N, Baillie J. Dieulafoy lesions: a review of 6 years of experience at a tertiary referral center. The American journal of gastroenterology. 2001 Jun 1;96(6):1688-94.
6. Malik A, Inayat F, Goraya MH, Almas T, Ishtiaq R, Malik S, Tarar ZI. Jejunal Dieulafoy's lesion: a systematic review of evaluation, diagnosis, and management. Journal of investigative medicine high impact case reports. 2021 Jan; 9:2324709620987703.
7. Wan Muhaizan WM, Julia MJ, Al Amin D. Calibre persistent submucosal artery of the jejunum: a rare cause of massive gastrointestinal bleeding. The Malaysian Journal of Pathology. 2002 Dec 1;24(2):113-6.
8. Chaer RA, Helton SW. Dieulafoy's disease. Journal of the American College of Surgeons. 2003 Feb 1;196(2):290-6.
9. Khan R, Mahmad A, Gobrial M, Onwochei F, Shah K. The diagnostic dilemma of Dieulafoy's lesion. Gastroenterology Research. 2015 Aug;8(3-4):201.
10. Kozan R, Gülen M, Yılmaz TU, Leventoğlu S, Yılmaz E. Massive lower gastrointestinal bleeding from a jejunal Dieulafoy lesion. Turkish Journal of Surgery/Ulusal cerrahi dergisi. 2014;30(4):225.
11. Yehya M, Mayovska O, Flick A, Moszczynski Z. A jejunal Dieulafoy lesion: rare case necessitating surgical intervention. International Journal of Surgery Case Reports. 2020 Jan 1; 72:541-5.
12. Schneider M, Höllerich J, Beyna T. Device-assisted enteroscopy: A review of available techniques and upcoming new technologies. World journal of gastroenterology. 2019 Jul 7;25(27):3538.
13. Saada M, Perek S, Agbaria M, Raz-Pasteur A. Massive gastrointestinal bleeding from a jejunal Dieulafoy lesion: an extraordinary presentation. Case Reports in Gastroenterology. 2019;13(3):508-13.