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Case report

Solitary jejunal cavernous hemangioma causing intermittent melena: A case report

Tae Gil Heo

Department of Surgery, Ilsan Paik Hospital, Inje University College of Medicine, Goyang, Republic of Korea

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ABSTRACT

Introduction and importance: Hemangioma of the small intestine is a rare disease that causes acute and chronic intestinal bleeding and is difficult to diagnose before surgery. This report presents a case of a cavernous hemangioma confined to the jejunum of a man with intermittent melena.

Case presentation: A 38-year-old man presented with an episode of melena one and a half month ago. He had similar symptoms a year ago and had undergone esophagogastroduodenoscopy, colonoscopy, and abdominopelvic computed tomography, but the results were normal. Abdominopelvic computed tomography performed after hospitalization showed a small intraluminal protruding mass in the jejunum. A jejunal hemangioma was found during laparoscopy and was successfully treated with intestinal resection.

Clinical discussion: It is difficult to identify the cause of gastrointestinal bleeding preoperatively in patients with normal esophagogastroduodenoscopy, colonoscopy, and abdominopelvic computed tomography results. Small bowel tumors, especially small hemangiomas, should be considered as a cause. Laparoscopy may be one option for finding and treating lesions of the small intestine in hospitals where capsule endoscopy or double-balloon intestinal enteroscopy is not available.

Conclusion: This report presents a case of a cavernous hemangioma localized in the jejunum of a 38-year-old man with intermittent melena that was successfully treated with laparoscopy and intestinal resection, thereby emphasizing the usefulness of laparoscopy in hospitals where capsule endoscopy or double-balloon intestinal enteroscopy is not available.

1. Introduction

Acute gastrointestinal (GI) bleeding is a common emergency and is a major cause of mortality. Esophageal gastroduodenal endoscopy and colonoscopy do not detect GI bleeding sources in approximately 3%–5% of patients. In such cases, most causative lesions are found in the small intestine [1].

Hemangioma of the small bowel is a rare disease that causes acute and chronic intestinal bleeding, and preoperative diagnosis is difficult in patients with normal esophagogastroduodenoscopy, colonoscopy, and abdominopelvic computed tomography (CT) findings. Emphasizing the usefulness of laparoscopy in hospitals where capsule endoscopy or double-balloon intestinal enteroscopy is not available, this report presents a case of a cavernous hemangioma confined to the jejunum of a 38-year-old man with intermittent melena that was successfully treated with laparoscopy and intestinal resection. This case report has been

reported in line with the SCARE criteria [2].

2. Presentation of case

The Institutional Review Board of our hospital approved this study and granted a waiver for informed consent owing to the retrospective nature of the case report.

A 38-year-old man presented with an episode of melena one and a half months ago in Malaysia. He was a Jehovah's Witness and a Korean resident in Malaysia and had no specific history including no previous abdominal surgery. He had similar symptoms a year ago and was examined at the Department of Gastroenterology. At that time, no abnormal findings were found on esophagogastroduodenoscopy, colonoscopy, and abdominopelvic CT, except for severe iron-deficiency anemia.

The patient was anxious about intestinal bleeding and was

Abbreviations: GI, gastrointestinal; CT, computed tomography.

^{*} Department of Surgery, Ilsan Paik Hospital, Inje University College of Medicine, #170, Juhwaro, Ilsanseogu, Goyangsi, Gyeonggido 10380, Republic of Korea. E-mail address: tgheo@paik.ac.kr.

hospitalized to determine the cause. His blood pressure, pulse rate, and respiratory rate were stable (140/70 mmHg; 55/min; and 36.7 $^{\circ}$ C, respectively). His laboratory blood tests were also normal without anemia at the time of admission. His peripheral blood smear showed no pathognomonic morphology. His blood biochemistry for anemia was normal except for serum iron [iron: 29 (normal range: 33–193) µg/dL].

Abdominopelvic CT performed after hospitalization revealed an intraluminal protruding mass in the jejunum. Contrast-enhanced axial (Fig. 1a) and coronal (Fig. 1b) abdominal CT showed a peripheral, nodular-enhancing, low-attenuated intraluminal protruding mass in the jejunum. There were no abnormal findings on the CT scan performed a year ago. Since capsule endoscopy could not be performed at the hospital, laparoscopy was performed after obtaining the patient's consent at the Department of Surgery.

During laparoscopy, which used three ports (a 10-mm camera port and two 5-mm working ports), a 1-cm angiomatous bluish-purple lesion with an abnormal vessel on the serosa of the jejunum was found. The small bowel lesion was easy to detect because of its shape and color (Fig. 2). The lesion was located 70 cm from the ligament of Treitz, and no other lesions were found. The abnormal segment of the jejunum was retrieved through an additional 3-cm incision in the umbilical port site. After small bowel resection with a margin of 3 cm on both sides, functional end-to-end anastomosis was performed.

Histopathological examination of the lesion showed a cavernous hemangioma. Pathological examination identified the neoplasm as a cavernous hemangioma of the small intestine with a pinkish-gray and soft section composed of congestive vascularized structures and fragile vascular veins (Fig. 3). The patient's postoperative course was uneventful. The patient was discharged on the 7th postoperative day. He decided to visit the surgical outpatient clinic when he returned to Korea after discharge, but he did not. At 3 years and 9 months after surgery, it was confirmed through e-mail that he has been healthy without other symptoms such as melena.

3. Discussion

Small intestine hemangioma is a rare congenital vascular hamartomatous tumor with single or multiple lesions, most commonly involving the jejunum and rarely the duodenum [3,4]. Hemangiomas, like other benign tumors of the GI tract that are found incidentally on clinical examination, can be difficult to diagnose early before surgery because they have no specific symptoms.

The most common presentation is chronic GI bleeding, which causes anemia of unknown cause and rarely leads to massive bleeding. Other rare presentations include intestinal obstruction, intussusception,

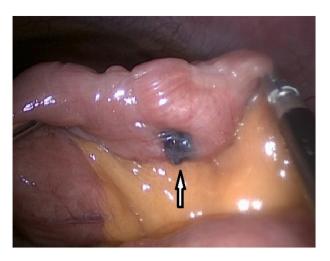


Fig. 2. Laparoscopic image showing the lesion (arrow).

intramural hematoma, perforation, and platelet sequestration [5]. As the symptoms of intestinal hemangioma are similar to those of other chronic bowel diseases, such as inflammatory bowel disease or tuberculosis, it is a differential diagnosis to be considered in these situations. Moreover, intestinal hemangiomas should be considered in all patients under investigation for anemia or GI bleeding.

Hemangiomas can be classified as spongy, which is the most frequent, capillary, or mixed, depending on the diameter of the injured blood vessel [5]. Tumor sizes can vary from a few millimeters to several centimeters (in giant tumors). In terms of gross features, the tumors are nodular, polypus, or diffusely infiltrative. The tumor is often localized to the submucosa due to the submucosal vascular plexus. Subserosal vasculature is rarely observed [5].

In cases of unclear GI bleeding, examinations such as CT, esophagogastroduodenoscopy, colonoscopy, or bone marrow puncture are essential, and capsule endoscopy, double-balloon intestinal enteroscopy, and multi-detector CT can be considered if necessary [6]. In cases of small hemangiomas, preoperative diagnosis is extremely difficult because they are rarely demonstrable with traditional techniques such as esophagoduodenoscopy and colonoscopy. However, several imaging modalities, including wireless capsule endoscopy, double-balloon intestinal enteroscopy, multiphase CT, and magnetic resonance imaging, are currently available for examining small bowel lesions [7]. The advent of capsule endoscopy and double-balloon intestinal enteroscopy allows a complete investigation of the small intestine and significantly

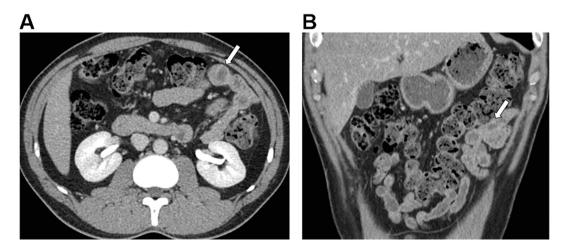


Fig. 1. Contrast-enhanced axial (a) and coronal (b) abdominal computed tomography scans show a peripheral, nodular-enhancing, low-attenuated intraluminal protruding mass (arrow) in the jejunum.

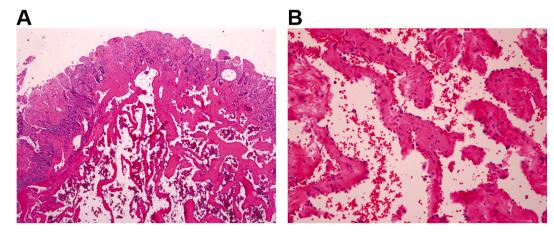


Fig. 3. a. Photomicrograph of the lesion. ×40: Submucosal tumor consists of large dilated vessels with variously thickened fibrous walls. The vessels are congested with blood. b. Photomicrograph of the lesion. ×200: The vessels are lined by the flattened endothelium.

improves the diagnosis of small intestinal lesions. Hemangiomas of the small intestine are an uncommon cause of GI bleeding and can be diagnosed using capsule endoscopy or double-balloon intestinal enteroscopy [8,9]. Small intestine capsule endoscopy is a non-invasive imaging test and may be recommended if the cause of the bleeding has not been identified after esophagogastroduodenoscopy and colonoscopy. In contrast, double-balloon intestinal enteroscopy is an invasive and highly sensitive diagnostic tool that allows both therapeutic and diagnostic interventions.

As a cavernous hemangioma of the small intestine is a rare condition without specific symptoms, early diagnosis can be difficult before surgery. Therefore, diagnostic examinations and treatments of this disease need to be further explored. The main treatment for hemangiomas is surgical resection of the affected area. Hemangiomas do not spread to lymph nodes or distant organs; therefore, local resection is sufficient.

The patient was a Jehovah's Witness. Jehovah's Witnesses refuse blood transfusions, which they consider a violation of God's law. Members are directed to refuse blood transfusions, even in "a life-ordeath situation." [10] Therefore, they are extremely sensitive to bleeding anywhere in the body and have a desire to eliminate the cause. He is a Korean residing in Malaysia, and he strongly hoped to treat the cause of intestinal bleeding, which caused melena. He refused blood transfusion under any circumstances. Since capsule endoscopy and double-balloon intestinal enteroscopy could not be performed in the hospital, the surgery was conducted under general anesthesia after obtaining consent for performing laparotomy using laparoscopic instruments.

Capsule endoscopy or double-balloon intestinal enteroscopy is required to identify small intestinal bleeding in patients with no abnormal findings on esophagogastroduodenoscopy or colonoscopy and no obvious bleeding lesions such as inflammatory small intestine disease or pronounced small intestine tumor on abdominopelvic CT. However, there are limited number of hospitals where capsule endoscopy or double-balloon intestinal enteroscopy is available. Thus, in hospitals where these procedures cannot be performed, even if the lesion cannot be found, laparoscopy may be the most optimal option as it allows simultaneous diagnosis and treatment. Fortunately, in the current case, the lesion was easily detected and treated through laparoscopy.

4. Conclusion

This report presents a case of a cavernous hemangioma localized in the jejunum of a 38-year-old man with intermittent melena, who was treated successfully with laparoscopy and intestinal resection, emphasizing the usefulness of laparoscopy in hospitals where capsule endoscopy or double-balloon intestinal enteroscopy is not available.

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Ethical approval

The Institutional Review Board of Inje University Ilsanpaik Hospital (ISPAIK 2021-05-004) approved this study.

Consent

The Institutional Review Board of Inje University Ilsanpaik Hospital granted a waiver for informed consent owing to the retrospective nature of the case report.

The consent for publication of this case report and accompanying images was obtained from the patient via e-mail. A copy of the consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Tae Gil Heo is the sole author of this article.

Registration of research studies

Not applicable.

Guarantor

Tae Gil Heo accepts the full responsibility for the article.

Provenance and peer review

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Declaration of competing interest

None.

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References

[1] H. Okazaki, Y. Fujiwara, S. Sugimori, Y. Nagami, N. Kameda, H. Machida, et al., Prevalence of mid-gastrointestinal bleeding in patients with acute overt

- gastrointestinal bleeding: multi-center experience with 1,044 consecutive patients, J. Gastroenterol. 44 (2009) 550–555.
- [2] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, for the SCARE Group, The SCARE 2020 guideline: updating consensus Surgical CAse REport (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230.
- [3] A.R. Ruiz, A.L. Ginsberg, Giant mesenteric hemangioma with small intestinal involvement: an unusual cause of recurrent gastrointestinal bleed and review of gastrointestinal hemangiomas, Dig. Dis. Sci. 44 (1999) 2545–2551.
- [4] A. Huber, A. Abdel Samie, D. Kychenko, L. Theilmann, A rare cause of recurrent iron-deficiency anemia: cavernous hemangioma of the small intestine, J. Gastrointestin. Liver Dis. 21 (2012) 343.
- [5] M. Pera, L. Márquez, J.M. Dedeu, J. Sánchez, M. Garcia, J.M. Ramón, et al., Solitary cavernous hemangioma of the small intestine as the cause of long-standing iron deficiency anemia, J. Gastrointest. Surg. 16 (2021) 2288–2290.
- [6] Y. Akazawa, K. Hiramatsu, T. Nosaka, Y. Saito, Y. Ozaki, K. Takahashi, et al., Preoperative diagnosis of cavernous hemangioma presenting with melena using

- wireless capsule endoscopy of the small intestine, Endosc. Int. Open 4 (2016) E249–E251.
- [7] I. Watari, S. Oka, S. Tanaka, M. Nakano, T. Aoyama, S. Yoshida, et al., Is occult obscure gastrointestinal bleeding a definite indication for capsule endoscopy? A retrospective analysis of diagnostic yield in patients with occult versus overt bleeding, Gastroenterol. Res. Pract. 2013 (9) (2013), 15463.
- [8] A. Magnano, A. Privitera, G. Calogero, G. Basile, G. Sanfilippo, Solitary hemangioma of the small intestine: an unusual cause of bleeding diagnosed at capsule endoscopy, J. Pediatr. Surg. 40 (2005) e25–e27.
- [9] M.N. de Mascarenhas-Saraiva, L.M. da Silva Araújo Lopes, Small-bowel tumors diagnosed by wireless capsule endoscopy: report of five cases, Endoscopy 35 (2003) 865–868.
- [10] D.T. Ridley, Jehovah's Witnesses' refusal of blood: obedience to scripture and religious conscience, J. Med. Ethics 25 (1999) 469–472.