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## Small intestinal hemangioma: Endoscopic or surgical intervention? A case report and review of literature

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### Abstract

#### BACKGROUND

Hemangioma of the small intestine is a rare vascular malformation. Before the advent of capsule endoscopy (CE) and balloon-assisted enteroscopy (BAE), preoperative diagnosis of this disease was extremely difficult.

#### CASE SUMMARY

In this study, we report a 24-year-old female with a large transmural small bowel cavernous hemangioma, which was diagnosed with CE and BAE preoperatively and removed successfully using minimally invasive surgery. Meanwhile, we perform a literature review of the studies about intestinal hemangiomas published after 2000. Literature review revealed that 91.9% of the lesions were diagnosed preoperatively by CE and/or BAE and 45.9% of them were treated endoscopically, which is a marked improvement compared to before 2000. Therefore, CE and BAE are useful modalities for the preoperative diagnosis of hemangiomas in the small intestine.

#### CONCLUSION

Endoscopic treatment of intestinal hemangioma is

generally prudent and might be suitable for multiple, relatively small lesions.

**Key words:** Hemangioma; Capsule endoscopy; Balloon-assisted enteroscopy; Endoscopic intervention; Surgery; Case report

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**Core tip:** Hemangioma of the small intestine is a rare disease and mostly presents as gastrointestinal bleeding. With the advent of capsule endoscopy and balloon-assisted enteroscopy, the preoperative diagnosis of this disease has been considerably improved. Surgical resection is the conventional treatment modality. With the improvement of endoscopic therapeutic interventions, less invasive procedures are becoming possible. However, potential risks of endoscopic treatment include bleeding and intestinal perforation. Since intestinal hemangiomas originate from the submucosal layer and some of them are transmural, endoscopic treatment might sometimes result in uncontrolled bleeding or perforation.

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## INTRODUCTION

Hemangioma of the small intestine is a rare disease, accounting for 7%-10% of all benign tumors of the small intestine<sup>[1,2]</sup>. It may be solitary or multiple, with the jejunum being the most common site of involvement<sup>[3]</sup>. The main presenting symptoms include hemorrhage, abdominal pain, obstruction, intussusceptions, or rarely, perforation<sup>[4,5]</sup>. It originates from the submucosal vascular plexuses and may extend into the muscular layer or beyond<sup>[6]</sup>. Histologically, hemangiomas are congenital benign vascular lesions that can be classified as capillary, cavernous, or mixed-type according to the size of the vascular channels<sup>[2]</sup>. With the advent of capsule endoscopy (CE) and balloon-assisted enteroscopy (BAE), complete investigation of the small bowel is possible<sup>[7]</sup>. The preoperative diagnosis of this disease has been considerably improved. Recent advances in endoscopic techniques have led to successful endoscopic intervention, but most large lesions have been treated surgically. Here, we present a case with solitary small bowel hemangioma, which was diagnosed preoperatively by CE and BAE and removed successfully using minimally invasive surgery.

## CASE PRESENTATION

### Chief complaints

A 24-year-old female suffered from recurrent melena and fatigue for 1 year.

### History of present illness

Over the past year, the patient experienced repeated black stool, accompanied by fatigue, without hematemesis, hematochezia, abdominal pain or fever. The lowest level of hemoglobin was 42 g/L.

### History of past illness

Past and family medical history was unremarkable.

### Physical examination

Physical examination showed moderate anemia. Detailed dermatological evaluation did not show any cutaneous lesions.

### Laboratory testing

Laboratory studies revealed moderated microcytic and hypochromic anemia (hemoglobin, 7.5 g/dL). Fecal occult blood test was positive.

### Imaging examination

Gastroscopy and colonoscopy were normal. CE was performed, showing a prominent polypoid lesion in the ileum with no sign of active bleeding (Figure 1). Transanal double-balloon enteroscopy (DBE) revealed a reddish purple lesion in the ileum about 80 cm proximal to the ileocecal valve (Figure 2A). A titanium clip was used to mark the limit reached. Transoral DBE was performed to assess the remainder of small bowel, which revealed no additional lesions (Figure 2B).

## MULTIDISCIPLINARY EXPERT CONSULTATION

### *Ping-Fang Hu, MD, Attending Doctor, Department of Gastroenterology*

From the endoscopic appearance of the lesion, it was most likely a hemangioma. Considering that the lesion was large and diffuse, endoscopic interventions such as endoscopic mucosal resection (EMR) and endoscopic sclerotherapy might lead to uncontrolled bleeding or perforation. Therefore, laparoscopic surgery was deemed the best choice.

### *Bin Shi, MD, Professor, Department of Gastroenterology*

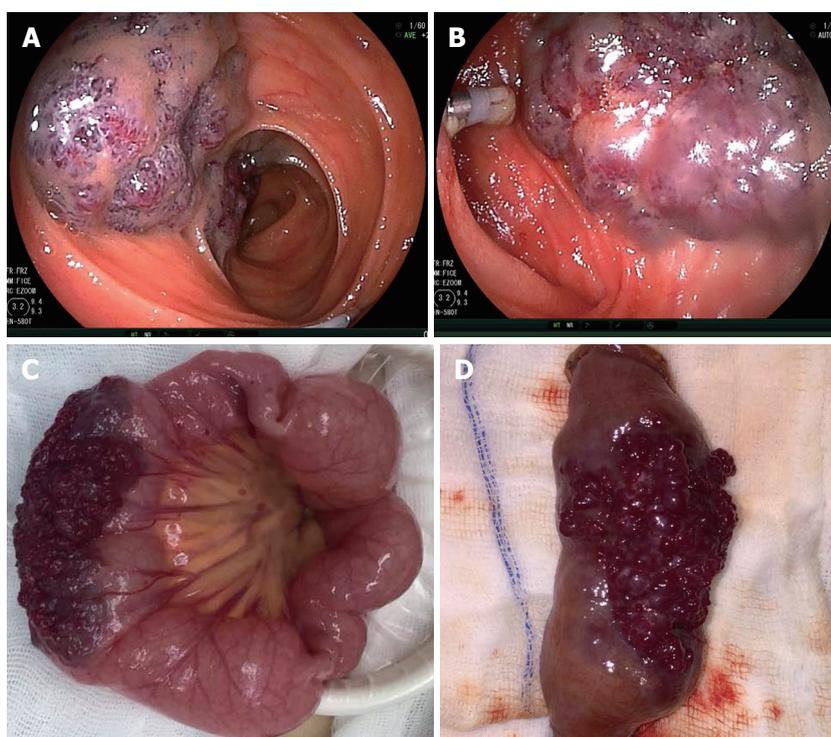
The patient had repeated bleeding and a large amount of bleeding every time. Since the lesion was large and diffuse, surgery would be better for the patient.

### *Han Chen, MD, Attending Doctor, Department of Surgery*

The patient suffered from recurrent melena in the past year. From the results of the CE and BAE, the cause



**Figure 1** Capsule endoscopic appearance of the lesion. Capsule endoscopy showed a prominent polypoid lesion in the ileum.



**Figure 2** Endoscopic and gross appearance of the lesion. A: Transanal double-balloon enteroscopy revealed a reddish purple lesion in the ileum about 80 cm proximal to the ileocecal valve, and a titanium clip was used to mark the limit reached; B: Transanal double-balloon enteroscopy showed the same lesion and the marked titanium clip; C: Gross intraoperative appearance of the lesion; D: Gross appearance of the lesion after resection.

is likely the small intestinal hemangioma. The surgical indication was explicit.

**Ning Su, MD, Attending Doctor, Department of Surgery**

The diagnosis is relatively clear. Since biopsy might lead to uncontrolled bleeding, we could not verify the diagnosis preoperatively.

**Wei-Jun Wang, Professor, Department of Surgery**

Imaging examination including ultrasound and CT scan did not find any abnormalities. From the endoscopic appearance of the lesion, it was most likely a hemangioma. The patient was a young female with a good health status. We could consider resecting the

lesion laparoscopically.

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## FINAL DIAGNOSIS

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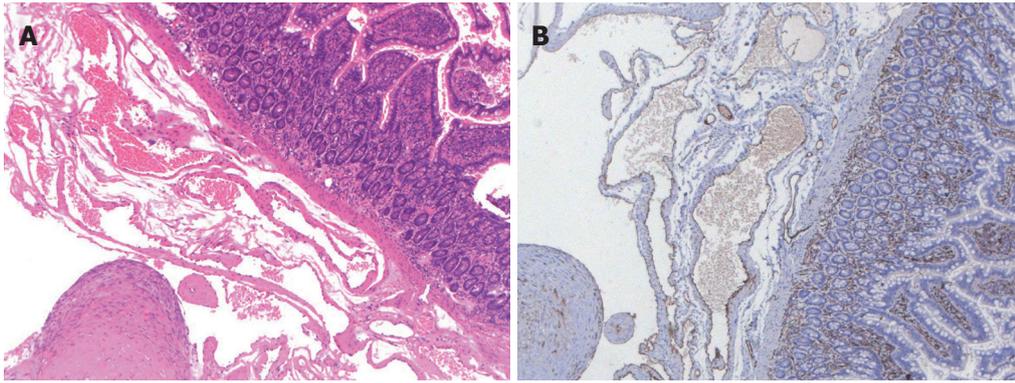
Small bowel bleeding and small intestinal hemangioma.

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## TREATMENT

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The patient was sent to laparoscopy, and a 5 cm × 3 cm × 3 cm purple-colored, raspberry-like lesion was found spreading diffusely along the serosal surface of the ileum (Figure 2C). The lesion was completely resected (Figure 2D). Hematoxylin-eosin staining (Figure 3A) and CD31 immunohistochemistry (Figure 3B) indicated



**Figure 3** Histopathological examination of the lesion. A: Hematoxylin-eosin staining showed a blood-filled sinus-like space in the whole layer of the ileum ( $\times 50$ ); B: Immunohistochemistry indicated the cells lined with the vascular spaces were CD31-positive ( $\times 50$ ).

a transmural cavernous hemangioma.

## OUTCOME AND FOLLOW-UP

The patient recovered quickly and had no further episodes of bleeding since the operation. The hemoglobin value increased to normal (12.4 g/dL) and was stable.

## DISCUSSION

Hemangioma accounts for only 0.05% of all gastrointestinal (GI) neoplasms. They mostly present with occult GI bleeding and iron deficiency anemia. Because of its rarity, it is not considered a common cause of GI bleeding. Previously, the preoperative diagnosis of this disease was difficult, and almost all cases were diagnosed during or after the operation<sup>[1]</sup>. With the introduction of CE and BAE over the past decades, the small intestine has now become an area that can be targeted<sup>[8]</sup>. We searched the PubMed database for studies about intestinal hemangiomas published after 2000 utilizing the following search terms: "hemangioma", "vascular malformation", "small intestine" and "small bowel". A manual search was also performed using the references of eligible articles. The language was limited to English. A total of 37 cases (16 women, 21 men, mean age 39 years) were retrieved and reviewed (Table 1). The most common manifestation included GI bleeding and anemia. A total of 75.7% (28/37) of the cases were single, and the common location of the small intestine was the jejunum (60.9%). Thirty-four of the 37 lesions (91.9%) were diagnosed before operation by CE and/or BAE. Of these cases, 11 were detected with CE alone, and 22 were diagnosed with both CE and BAE. Compared with the cases reported before 2000, a markedly increased proportion of cases were preoperatively diagnosed<sup>[1]</sup>. As in our case, CE was used to initially examine the GI tract, which was based on the algorithms for the diagnosis and treatment of obscure GI bleeding<sup>[7]</sup>. Both transanal DBE and transoral DBE were then performed to complete total enteroscopy, which was useful to localize the lesion and rule out

other lesions.

Surgical resection, which is relatively more invasive, is the conventional treatment modality for intestinal hemangiomas. With the improvement of endoscopic therapeutic interventions, less invasive procedures are becoming more widely employed. Of the 37 cases of intestinal hemangiomas published after 2000 (Table 1), 17 cases (45.9%) were treated endoscopically. Among them, 3 cases were removed by EMR, one case was treated by argon plasma coagulation, and 13 cases were subjected to sclerotherapy. Most of these lesions were multiple (14/17, 82.4%), and the lesions were relatively small. As suggested by the guideline on the management of small bowel bleeding, the patient should be managed with endoscopic therapy if a source of bleeding is found. Surgical treatment is generally regarded as a last resort<sup>[7]</sup>. Compared with surgery, endoscopic treatments including sclerotherapy and EMR are less invasive. However, they increase the potential risks of GI bleeding and intestinal perforation. Since intestinal hemangiomas originate from the submucosal layer, endoscopic treatment such as EMR is dangerous because of the risk of perforation. Endoscopic treatment might lead to perforation because some intestinal hemangiomas were transmural, as in our case. Considering that the hemangioma was large in the current case, uncontrolled bleeding would probably occur after endoscopic intervention. After discussion with a multidisciplinary team, which included gastroenterologists, endoscopists and surgeons, we decided to remove the lesion by laparoscopy. It turned out that a laparoscopic approach was likely the best choice for our case, as the lesion was relatively large and most importantly, transmural. Thus, endoscopic treatment of intestinal hemangioma should be prudent. It is likely suitable for multiple, relatively small lesions.

In conclusion, we present a case of small bowel hemangioma that was preoperatively diagnosed by CE and BAE and treated by laparoscopy. We believe it is important for both the endoscopist and surgeons to recognize this somewhat unusual lesion. It is recommended that careful consideration of the indications for

**Table 1 Summary of hemangioma of small intestine reported after 2000**

Ref.	Country	Case	Sex/age	Complaint	Diagnosis	Location	Single/multiple	Treatment	Pathology
Easler <i>et al</i> <sup>[9]</sup>	United States	1	M/71	Anemia, melena	BAE	Jejunum	Single	EMR	Cavernous
Ng <i>et al</i> <sup>[10]</sup>	China	1	F/20	Anemia	Small bowel enema	Terminal ileum	Multiple	APC	-
Wardi <i>et al</i> <sup>[11]</sup>	Israel	1	M/77	Anemia, melena	CE	Ileum	Single	Laparoscopy	Capillary
Ersoy <i>et al</i> <sup>[6]</sup>	Turkey	1	F/50	Melena, hematemesis	CE + BAE	Proximal jejunum	Single	Laparoscopy	Cavernous
Fernandes <i>et al</i> <sup>[12]</sup>	Portugal	1	F/56	Hematochezia, syncope	CE	Ileum	Single	Laparoscopy	Cavernous
Law <sup>[13]</sup>	China	1	F/31	Melena	CE + BAE	Jejunum	Single	Laparoscopy	Cavernous
Ning <i>et al</i> <sup>[4]</sup>	China	1	M/10	Melena	BAE	Jejunum/ileum	Multiple	Polidocanol injection	-
Elias <i>et al</i> <sup>[15]</sup>	United States	1	M/30	Anemia	CE + BAE	Jejunum	Multiple	Surgery	Cavernous
Shibuya <i>et al</i> <sup>[16]</sup>	Japan	1	M/74	Melena	CE + BAE	jejunum	Single	EMR	Capillary
Willert <i>et al</i> <sup>[6]</sup>	Australia	1	M/19	Anemia	CE + BAE	Jejunum/ileum	Multiple	EMR	Cavernous
Igawa <i>et al</i> <sup>[17]</sup>	Japan	12	6M/6F	Gastrointestinal bleeding	CE + BAE	Jejunum/ileum	7 single/5 multiple	Polidocanol injection	-
Takase <i>et al</i> <sup>[18]</sup>	Japan	2	F-62/M-52	Melena	CE + BAE	Jejunum/ileum	Single	Laparoscopy	Cavernous/capillary
Akazawa <i>et al</i> <sup>[19]</sup>	Japan	1	F/56	Melena	CE + BAE	Jejunum	Single	Laparoscopy	Cavernous
Chen <i>et al</i> <sup>[20]</sup>	United States	1	M/23	Fatigue	CE	Ileum	Single	Laparoscopy	Cavernous
Dhumane <i>et al</i> <sup>[21]</sup>	France	1	M/60	Anemia	CE + BAE	Jejunum	Single	Laparoscopy	Cavernous
Bae <i>et al</i> <sup>[22]</sup>	South Korea	1	M/13	Dizziness, fatigue	CE	Jejunum	Single	Laparoscopy	Cavernous
Huber <i>et al</i> <sup>[23]</sup>	Germany	1	M/23	Weakness, dizziness	CE + BAE	Jejunum	Single	Laparoscopy	Cavernous
Quentin <i>et al</i> <sup>[5]</sup>	France	1	F/32	Hematochezia	CE	Jejunum	Single	Laparoscopy	Cavernous
Khurana <i>et al</i> <sup>[24]</sup>	United States	1	M/62	Melena	BAE	Jejunum	Single	Surgery	Cavernous
Pera <i>et al</i> <sup>[25]</sup>	Spain	1	M/16	Fatigue	CE	Jejunum	Single	Laparoscopy	-
Pinho <i>et al</i> <sup>[26]</sup>	Portugal	1	F/9	Melena, anemia	CE	Ileum	Single	Surgery	Cavernous
Magnano <i>et al</i> <sup>[27]</sup>	Italy	1	M/13	Fatigue, malaise	CE	Ileum	Single	Laparoscopy	Cavernous
Kuo <i>et al</i> <sup>[28]</sup>	China	1	F/20	Abdominal pain	-	Jejunum	Single	Laparoscopy	Cavernous
Guardiola <i>et al</i> <sup>[29]</sup>	Spain	1	M/19	Anemia	CE	Ileum	Single	Laparoscopy	Cavernous
Purdy-Payne <i>et al</i> <sup>[30]</sup>	United States	1	F/20	Abdominal pain	-	Terminal ileum	Single	Laparoscopy	Cavernous

CE: Capsule endoscopy; BAE: Balloon assisted enteroscopy; EMR: Endoscopic mucosal resection; APC: Argon plasma coagulation.

endoscopic treatment. As in our case, hemangiomas may sometimes involve the entire wall of the intestine. Endoscopic intervention may lead to uncontrolled bleeding or perforation. For the large and diffuse lesions, a laparoscopic excision might be a better approach.

## EXPERIENCES AND LESSONS

Hemangioma of the small intestine is a rare disease, which mostly presented as occult GI bleeding and iron deficiency anemia. With the advent of CE and BAE, the diagnosis of lesions in the small intestine has been considerably improved. Endoscopic treatment of intestinal hemangioma should be prudent, and it might be suitable for multiple and relatively small lesions.

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