**Name of Journal:** *World Journal of Gastrointestinal Oncology*

**Manuscript NO:** 42247

**Manuscript Type:** CASE REPORT

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

Hu PF *et al*. Small intestinal hemangioma

Ping-Fang Hu, Han Chen, Xiao-Hang Wang, Wei-Jun Wang, Ning Su, Bin Shi

**Ping-Fang Hu, Xiao-Hang Wang, Bin Shi,** Department of Gastroenterology, Changzheng Hospital, Second Military Medical University, Shanghai 200003, China

**Han Chen,** Department of General Surgery, Hongkou Branch of Changhai Hospital, Second Military Medical University, Shanghai 200081, China

**Wei-Jun Wang, Ning Su,** Department of General Surgery, Changzheng Hospital, Second Military Medical University, Shanghai 200003, China

**ORCID number:** Ping-Fang Hu (0000-0002-2790-9674); Han Chen (0000-0002-5912-3692); Xiao-Hang Wang (0000-0001-7598-9755); Wei-Jun Wang ([0000-0002-8831-0665](http://orcid.org/0000-0002-8831-0665)); Ning Su (0000-0002-8423-0975); Bin Shi (0000-0003-1775-4581).

**Author contributions:** Hu PF and Chen H reviewed literatures, designed the case reports presentation and wrote manuscript; Wang XH participated in manuscript preparation, revision, patient’s investigation and treatment; Wang WJ and Su N participated in patients’ investigation and treatment and provided the gross and pathology images; Shi B designed the case reports presentation and revised the manuscript.

**Informed consent statement:** The study participant provided informed written consent prior to study enrollment.

**Conflict of interest statement:** The authors have no conflict of interest to declare.

**CARE Checklist (2016) statement:** The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

**Open-Access:** This article is an open-access article which was selected by an in-house editor and fully peer-reviewed by external reviewers. It is distributed in accordance with the Creative Commons Attribution Non Commercial (CC BY-NC 4.0) license, which permits others to distribute, remix, adapt, build upon this work non-commercially, and license their derivative works on different terms, provided the original work is properly cited and the use is non-commercial. See: http://creativecommons.org/licenses/by-nc/4.0/

**Manuscript source:** Unsolicited manuscript

**Correspondence to: Bin Shi, MD, Professor,** Department of Gastroenterology, Changzheng Hospital, Second Military Medical University, 415 Fengyang Road, Shanghai 200003, China. [shibin7305@smmu.edu.cn](mailto:shibin7305@smmu.edu.cn)

**Telephone:** +86-21-81885346

**Fax:** +86-21-81886924

**Received:** September 18, 2018

**Peer-review started:** September 18, 2018

**First decision:** October 15, 2018

**Revised:** October 24, 2018

**Accepted:** November 7, 2018

**Article in press:**

**Published online:**

**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 is 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 were markedly improved as compared with that before 2000. Therefore, CE and BAE are useful modalities for preoperative diagnosis of hemangiomas in the small intestine.

***CONCLUSION***

Endoscopic treatment of intestinal hemangioma should be prudent, which might be suitable for lesions which are multiple and relatively small.

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

© **The Author(s) 2018.** Published by Baishideng Publishing Group Inc. All rights reserved.

**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.

Hu PF, Chen H, Wang XH, Wang WJ, Su N, Shi B. Small intestinal hemangioma: endoscopic or surgical intervention? A case report and review of literature. *World J Gastrointest Oncol* 2018; In press

**INTRODUCTION**

Hemangioma of the small intestine is a rare disease, accounting for 7%-10% of all benign tumors of the small intestine[[1](#_ENREF_1),[2](#_ENREF_2)]. It may be solitary or multiple, with jejunum being the most common site of involvement[[3](#_ENREF_3)]. The main presenting symptoms include hemorrhage, abdominal pain, obstruction, intussusceptions, or rarely perforation[[4](#_ENREF_4),[5](#_ENREF_5)]. It originates from the submucosal vascular plexuses, and may extend into the muscular layer or beyond[[6](#_ENREF_6)]. 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[[2](#_ENREF_2)]. With the advent of capsule endoscopy (CE) and balloon-assisted enteroscopy (BAE), complete investigation of the small bowel is possible[[7](#_ENREF_7)]. And 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 one 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). Transannal 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 small bowel, which appreciated 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 hemangioma. Considering 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 would be 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 patients.

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

The patients suffered from recurrent melena in the past year. From the results of the CE and BAE, the cause might probably be 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 abnormal. From the endoscopic appearance of the lesion, it was most likely hemangioma. The patient was a young lady with a good health status. We could consider resecting the lesion laparoscopically.

**FINAL DIAGNOSIS**

Small bowel bleeding and small intestinal hemangioma.

**TREATMENT**

The patient was sent to laparoscopy and a 5 cm × 3 cm × 3 cmpurple-colored, raspberry-like lesion was found spreading diffusely along the serosal surface of the ileum (Figure 2C). The lesion was resected completely (Figure 2D). Hematoxylin-eosin staining (Figure 3A) and CD31 immunohistochemistry (Figure 3B) indicated 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 presented with occult GI bleeding and iron deficiency anemia. Because of its rarity, it is not considered as the common cause of GI bleeding. Previously, the preoperative diagnosis of this disease was difficult and almost all cases were diagnosed during or after operation[[1](#_ENREF_1)]. With the introduction of CE and BAE over the past decades, the small intestine has now become an area which can be targeted[[8](#_ENREF_8)]. We searched the PubMed database for the studies about intestinal hemangiomas published after 2000, utilizing the following search terms “hemangioma”, “vascular malformation”, “small intestine” and “small bowel”. And a manual search was also carried out using references of eligible articles. 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. 75.7% (28/37) of the cases are 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 of them. Compared with the cases reported before 2000, a markedly increased proportion of cases were diagnosed preoperatively[[1](#_ENREF_1)]. As in our case, CE was used to examine the GI tract initially, which was based on the algorithms for the diagnosis and treatment of obscure GI bleeding[[7](#_ENREF_7)]. And 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 mean of 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, patient should be managed with endoscopic therapy if a source of bleeding is found. And surgical treatment is generally regarded as a last resort[[7](#_ENREF_7)]. 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. And endoscopic treatment might lead to perforation because some intestinal hemangiomas were transmural, as in our case. Considering the hemangioma was large in the current case, uncontrolled bleeding would probably occur after endoscopic intervention. After discussion with a multiple disciplinary team which included gastroenterologists, endoscopists and surgeons, we decided to remove the lesion by laparoscopy. It turned out that a laparoscopic approach might be 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. And it might be suitable for the lesions which are multiple and relatively small.

In conclusion, we present here a case with small bowel hemangioma which was diagnosed by CE and BAE preoperatively and treated by laparoscopy. We believe it is important for both the endoscopist and surgeons to recognize this somewhat unusual lesion. And it is recommended careful consideration of the indications of endoscopic treatment. As in our case, hemangiomas may sometimes involve the entire wall of the intestine. And 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 the lesions which are multiple and relatively small.

**REFERENCES**

1 **Ramanujam PS**, Venkatesh KS, Bettinger L, Hayashi JT, Rothman MC, Fietz MJ. Hemangioma of the small intestine: case report and literature review. *Am J Gastroenterol* 1995; **90**: 2063-2064 [PMID: 7485031]

2 **Kumar N**, Adam SZ, Goodhartz LA, Hoff FL, Lo AA, Miller FH. Beyond hepatic hemangiomas: the diverse appearances of gastrointestinal and genitourinary hemangiomas. *Abdom Imaging* 2015; **40**: 3313-3329 [PMID: 26239397 DOI: 10.1007/s00261-015-0515-8]

3 **Quentin V**, Lermite E, Lebigot J, Marinnes MZ, Arnaud JP, Boyer J. Small bowel cavernous hemangioma: wireless capsule endoscopy diagnosis of a surgical case. *Gastrointest Endosc* 2007; **65**: 550-552 [PMID: 17321267 DOI: 10.1016/j.gie.2006.12.024]

4 **Rao AB**, Pence J, Mirkin DL. Diffuse infantile hemangiomatosis of the ileum presenting with multiple perforations: a case report and review of the literature. *J Pediatr Surg* 2010; **45**: 1890-1892 [PMID: 20850639 DOI: 10.1016/j.jpedsurg.2010.05.019]

5 **Ruiz AR Jr**, Ginsberg AL. Giant mesenteric hemangioma with small intestinal involvement: an unusual cause of recurrent gastrointestinal bleed and review of gastrointestinal hemangiomas. *Dig Dis Sci* 1999; **44**: 2545-2551 [PMID: 10630511 DOI: 10.1023/A:1026659710815]

6 **Ersoy O**, Akin E, Demirezer A, Koseoglu H, Balci S, Kiyak G. Cavernous haemangioma of small intestine mimicking gastrointestinal stromal tumour. *Arab J Gastroenterol* 2013; **14**: 139-140 [PMID: 24206746 DOI: 10.1016/j.ajg.2013.08.008]

7 **Gerson LB**, Fidler JL, Cave DR, Leighton JA. ACG Clinical Guideline: Diagnosis and Management of Small Bowel Bleeding. *Am J Gastroenterol* 2015; **110**: 1265-87; quiz 1288 [PMID: 26303132 DOI: 10.1038/ajg.2015.246]

8 **Willert RP**, Chong AK. Multiple cavernous hemangiomas with iron deficiency anemia successfully treated with double-balloon enteroscopy. *Gastrointest Endosc* 2008; **67**: 765-767 [PMID: 18155208 DOI: 10.1016/j.gie.2007.07.044]

9 **Easler JJ**, Papachristou GI. A case of obscure gastrointestinal bleeding. *Gastroenterology* 2012; **142**: 700, 1044 [PMID: 22370215 DOI: 10.1053/j.gastro.2011.09.009]

10 **Ng EK**, Cheung FK, Chiu PW. Blue rubber bleb nevus syndrome: treatment of multiple gastrointestinal hemangiomas with argon plasma coagulator. *Dig Endosc* 2009; **21**: 40-42 [PMID: 19691801 DOI: 10.1111/j.1443-1661.2008.00817.x]

11 **Wardi J**, Shahmurov M, Czerniak A, Avni Y. Clinical challenges and images in GI. Capillary hemangioma of small intestine. *Gastroenterology* 2007; **132**: 1656, 2084 [PMID: 17484862 DOI: 10.1053/j.gastro.2007.03.081]

12 **Fernandes D**, Dionísio I, Neves S, Duarte P. Cavernous hemangioma of small bowel: a rare cause of digestive hemorrhage. *Rev Esp Enferm Dig* 2014; **106**: 214-215 [PMID: 25007019]

13 **Law WL**. Cavernous hemangioma: uncommon cause of obscure gastrointestinal bleeding. *J Am Coll Surg* 2007; **205**: 511 [PMID: 17765169 DOI: 10.1016/j.jamcollsurg.2006.10.035]

14 **Ning S**, Zhang Y, Zu Z, Mao X, Mao G. Enteroscopic sclerotherapy in blue rubber bleb nevus syndrome. *Pak J Med Sci* 2015; **31**: 226-228 [PMID: 25878650 DOI: 10.12669/pjms.311.5858]

15 **Elias G**, Toubia N. Hemangioma of the small intestine presenting with recurrent overt, obscure gastrointestinal bleeding. *Clin Gastroenterol Hepatol* 2010; **8**: A18, A18.e1 [PMID: 19362610 DOI: 10.1016/j.cgh.2009.03.036]

16 **Shibuya T**, Osada T, Mitomi H, Takeda T, Nomura O, Nakayama H, Hidaka Y, Mori H, Beppu K, Sakamoto N, Nagahara A, Otaka M, Ogihara T, Yao T, Watanabe S. Jejunal capillary hemangioma treated by using double-balloon endoscopy (with video). *Gastrointest Endosc* 2010; **72**: 660-661 [PMID: 20546731 DOI: 10.1016/j.gie.2009.12.051]

17 **Igawa A**, Oka S, Tanaka S, Kunihara S, Nakano M, Chayama K. Polidocanol injection therapy for small-bowel hemangioma by using double-balloon endoscopy. *Gastrointest Endosc* 2016; **84**: 163-167 [PMID: 26907744 DOI: 10.1016/j.gie.2016.02.021]

18 **Takase N**, Fukui K, Tani T, Nishimura T, Tanaka T, Harada N, Ueno K, Takamatsu M, Nishizawa A, Okamura A, Kaneda K. Preoperative detection and localization of small bowel hemangioma: Two case reports. *World J Gastroenterol* 2017; **23**: 3752-3757 [PMID: 28611528 DOI: 10.3748/wjg.v23.i20.3752]

19 **Akazawa Y**, Hiramatsu K, Nosaka T, Saito Y, Ozaki Y, Takahashi K, Naito T, Ofuji K, Matsuda H, Ohtani M, Nemoto T, Suto H, Yamaguchi A, Imamura Y, Nakamoto Y. Preoperative diagnosis of cavernous hemangioma presenting with melena using wireless capsule endoscopy of the small intestine. *Endosc Int Open* 2016; **4**: E249-E251 [PMID: 27004239 DOI: 10.1055/s-0041-111321]

20 **Chen CH**, Jones J, McGowan P. Profound iron deficiency anemia caused by a small-intestinal cavernous hemangioma. *Gastrointest Endosc* 2009; **69**: 1392-3; discussion 1393 [PMID: 19481664 DOI: 10.1016/j.gie.2009.01.049]

21 **Dhumane P**, Mutter D, D'Agostino J, Mavrogenis G, Leroy J, Marescaux J. Small bowel exploration and resection using single-port surgery: a safe and feasible approach. *Colorectal Dis* 2013; **15**: 109-114 [PMID: 22672499 DOI: 10.1111/j.1463-1318.2012.03118.x]

22 **Bae SJ**, Hwang G, Kang HS, Song HJ, Chang WY, Maeng YH, Kang KS. Single Cavernous Hemangioma of the Small Bowel Diagnosed by Using Capsule Endoscopy in a Child with Chronic Iron-Deficiency Anemia. *Clin Endosc* 2015; **48**: 340-344 [PMID: 26240811 DOI: 10.5946/ce.2015.48.4.340]

23 **Huber A**, Abdel Samie A, Kychenko D, Theilmann L. A rare cause of recurrent iron-deficiency anemia: cavernous hemangioma of the small intestine. *J Gastrointestin Liver Dis* 2012; **21**: 343 [PMID: 23256111]

24 **Khurana V**, Dala R, Barkin JS. Small bowel cavernous hemangioma. *Gastrointest Endosc* 2004; **60**: 96 [PMID: 15229433 DOI: 10.1016/S0016-5107(04)01292-1]

25 **Pera M**, Márquez L, Dedeu JM, Sánchez J, Garcia M, Ramón JM, Puigvehí M. Solitary cavernous hemangioma of the small intestine as the cause of long-standing iron deficiency anemia. *J Gastrointest Surg* 2012; **16**: 2288-2290 [PMID: 22875598 DOI: 10.1007/s11605-012-1991-6]

26 **Pinho R**, Rodrigues A, Proença L, Silva AP, Fernandes S, Leite S, Amaral I, de Sousa P, Fraga J. Solitary hemangioma of the small bowel disclosed by wireless capsule endoscopy. *Gastroenterol Clin Biol* 2008; **32**: 15-18 [PMID: 18405648 DOI: 10.1016/j.gcb.2007.11.004]

27 **Magnano A**, Privitera A, Calogero G, Nanfito' L, Basile G, Sanfilippo G. Solitary hemangioma of the small intestine: an unusual cause of bleeding diagnosed at capsule endoscopy. *J Pediatr Surg* 2005; **40**: e25-e27 [PMID: 16226971 DOI: 10.1016/j.jpedsurg.2005.06.014]

28 **Kuo LW**, Chuang HW, Chen YC. Small bowel cavernous hemangioma complicated with intussusception: report of an extremely rare case and review of literature. *Indian J Surg* 2015; **77**: 123-124 [PMID: 25972669 DOI: 10.1007/s12262-014-1194-3]

29 **Guardiola A**, Navajas J, Valle J, López-Pardo R, Rodríguez-Merlo R, Lombera Mdel M, Alcántara M. Small bowel giant cavernous hemangioma diagnosed by capsule endoscopy. *Rev Esp Enferm Dig* 2012; **104**: 277-278 [PMID: 22662783 DOI: 10.4321/S1130-01082012000500011]

30 **Purdy-Payne EK**, Miner JF, Foles B, Tran TA. The "Endothelialized Muscularis Mucosae": A Case Report Describing a Large Cavernous Hemangioma at the Terminal Ileum and a New Histologic Clue for Preoperative Diagnosis from Endoscopic Biopsy. *Case Rep Gastrointest Med* 2015; **2015**: 454836 [PMID: 26442160 DOI: 10.1155/2015/454836]

**P-Reviewer:** Cao ZF, Sawaki A, Yarema RR **S-Editor:** Ji FF **L-Editor: E-Editor:**

**Specialty type:** Oncology

**Country of origin:** China

**Peer-review report classification**

Grade A (Excellent): 0

Grade B (Very good): 0

Grade C (Good): C, C, C

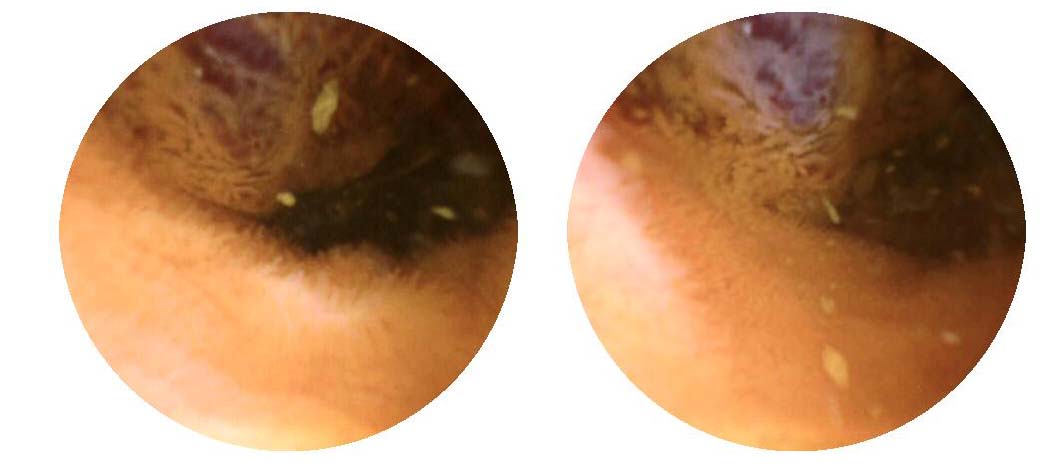
Grade D (Fair): 0

Grade E (Poor): 0

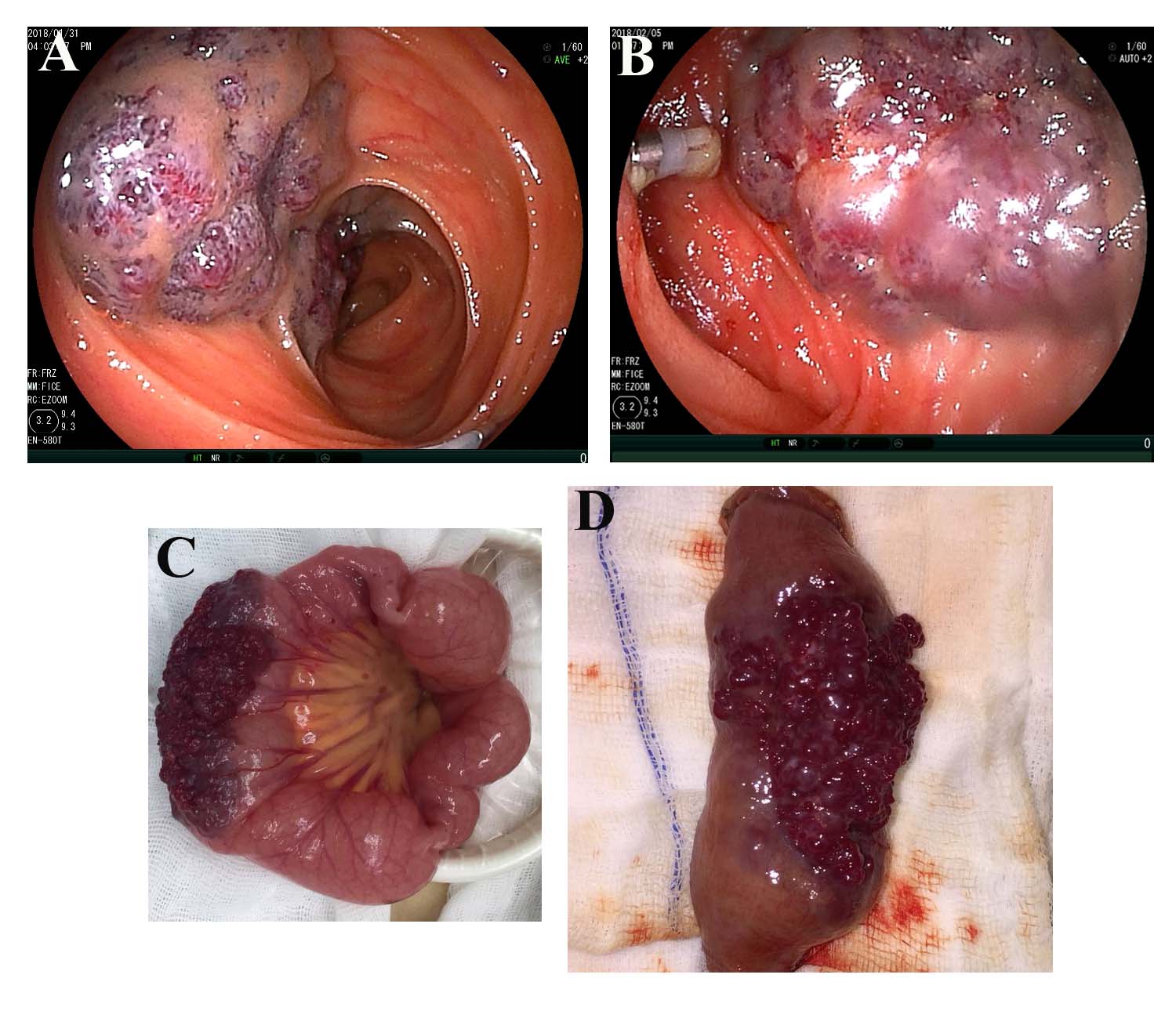
**Table 1 Summary of hemangioma of small intestine reported after 2000**

|  |  |  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- | --- | --- |
| **Ref.** | **Country** | **Case** | **Sex/age** | **Complaint** | **Diagnosis** | **Location** | **Single/multiple** | **Treatment** | **Pathology** |
| Easler *et al*[[9](#_ENREF_9)] | United States | 1 | M/71 | Anemia, melena | BAE | Jejunum | Single | EMR | Cavernous |
| Ng *et al*[[10](#_ENREF_10)] | China | 1 | F/20 | Anemia | Small bowel enema | Terminal ileum | Multiple | APC | - |
| Wardi *et al*[[11](#_ENREF_11)] | Israel | 1 | M/77 | Anemia, melena | CE | Ileum | Single | Laparoscopy | Capillary |
| Ersoy *et al*[[6](#_ENREF_6)] | Turkey | 1 | F/50 | Melena, haematemesis | CE + BAE | Proximal jejunum | Single | Laparoscopy | Cavernous |
| Fernandes *et al*[[12](#_ENREF_12)] | Portugal | 1 | F/56 | Hematochezia, syncope | CE | Ileum | Single | Laparoscopy | Cavernous |
| Law[[13](#_ENREF_13)] | China | 1 | F/31 | Melena | CE+ BAE | Jejunum | Single | Laparoscopy | Cavernous |
| Ning *et al*[[14](#_ENREF_14)] | China | 1 | M/10 | Melena | BAE | Jejunum/ileum | Multiple | Polidocanol injection | - |
| Elias *et al*[[15](#_ENREF_15)] | United States | 1 | M/30 | Anemia | CE+ BAE | Jejunum | Multiple | Surgery | Cavernous |
| Shibuya *et al*[[16](#_ENREF_16)] | Japan | 1 | M/74 | Melena | CE+ BAE | jejunum | Single | EMR | Capillary |
| Willert *et al*[[8](#_ENREF_8)] | Australia | 1 | M/19 | Anemia | CE+ BAE | Jejunum/ileum | Multiple | EMR | Cavernous |
| Igawa *et al*[[17](#_ENREF_17)] | Japan | 12 | 6M/6F | Gastrointestinal bleeding | CE+BAE | Jejunum/ileum | 7 single/5 multiple | Polidocanol injection | - |
| Takase *et al*[[18](#_ENREF_18)] | Japan | 2 | F-62/M-52 | Melena | CE+ BAE | Jejunum/ileum | Single | Laparoscopy | Cavernous/capillary |
| Akazawa *et al*[[19](#_ENREF_19)] | Japan | 1 | F/56 | Melena | CE + BAE | Jejunum | Single | Laparoscopy | Cavernous |
| Chen *et al*[[20](#_ENREF_20)] | United States | 1 | M/23 | Fatigue | CE | Ileum | Single | Laparoscopy | Cavernous |
| Dhumane *et al*[[21](#_ENREF_21)] | France | 1 | M/60 | Anemia | CE + BAE | Jejunum | Single | Laparoscopy | Cavernous |
| Bae *et al*[[22](#_ENREF_22)] | South Korea | 1 | M/13 | Dizziness, fatigue | CE | Jejunum | Single | Laparoscopy | Cavernous |
| Huber *et al*[[23](#_ENREF_23)] | Germany | 1 | M/23 | Weakness, dizziness | CE + BAE | Jejunum | Single | Laparoscopy | Cavernous |
| Quentin *et al*[[3](#_ENREF_3)] | France | 1 | F/32 | Hematochezia | CE | Jejunum | Single | Laparoscopy | Cavernous |
| Khurana *et al*[[24](#_ENREF_24)] | United States | 1 | M/62 | Melena | BAE | Jejunum | Single | Surgery | Cavernous |
| Pera *et al*[[25](#_ENREF_25)] | Spain | 1 | M/16 | Fatigue | CE | Jejunum | Single | Laparoscopy | - |
| Pinho *et al*[[26](#_ENREF_26)] | Portugal | 1 | F/9 | Melena, anemia | CE | Ileum | Single | Surgery | Cavernous |
| Magnano *et al*[[27](#_ENREF_27)] | Italy | 1 | M/13 | Fatigue, malaise | CE | Ileum | Single | Laparoscopy | Cavernous |
| Kuo *et al*[[28](#_ENREF_28)] | China | 1 | F/20 | Abdominal pain | - | Jejunum | Single | Laparoscopy | Cavernous |
| Guardiola *et al*[[29](#_ENREF_29)] | Spain | 1 | M/19 | Anemia | CE | Ileum | Single | Laparoscopy | Cavernous |
| Purdy-Payne *et al*[[30](#_ENREF_30)] | United States | 1 | F/20 | Abdominal pain | - | Terminal ileum | Single | Laparoscopy | Cavernous |

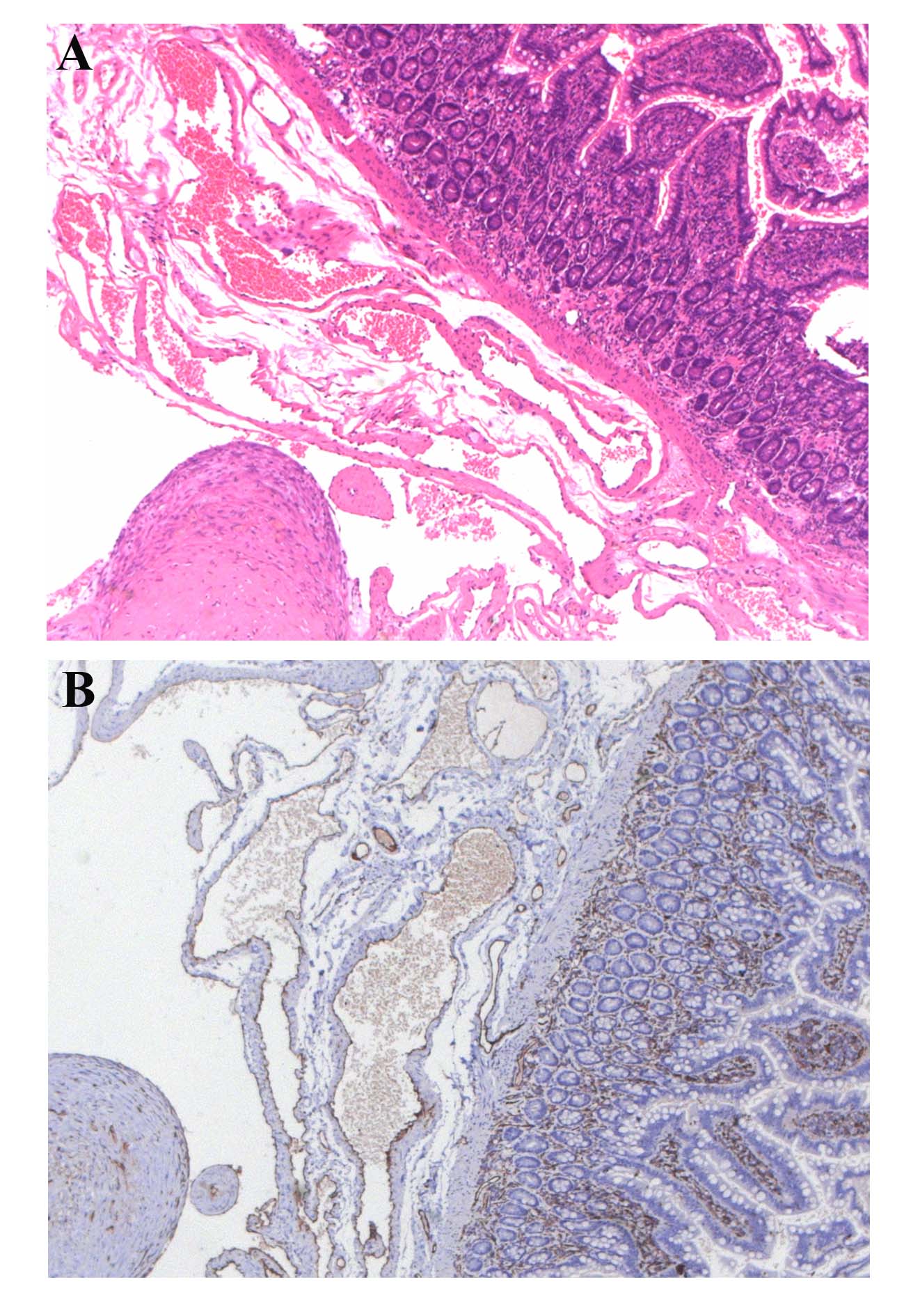
CE: Capsule endoscopy; BAE: Balloon assisted enteroscopy; EMR: Endoscopic mucosal resection; APC: Argon plasma coagulation.



**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: Transannal 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: transoral 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 resected.



**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 (× 50); B: Immunohistochemistry indicated the cells lined with the vascular spaces were CD31-positive (× 50).