Preoperative diagnosis of cavernous hemangioma presenting with melena using wireless capsule endoscopy of the small intestine

Background and study aims: Primary neoplasms of the small intestine are relatively rare in all age groups, accounting for about 5 % of all gastrointestinal tumors [1]. Cavernous hemangiomas of the small intestine are also rare, can cause gastrointestinal bleeding, and are extremely difficult to diagnose preoperatively [2]. We present a patient who presented with melena and iron deficiency anemia, for whom wireless capsule endoscopy and single-balloon enteroscopy facilitated the diagnosis of cavernous hemangioma.

Introduction

Cavernous hemangioma of the small intestine is a rare disease. Because hemangiomas can cause massive gastrointestinal bleeding, emergency surgery may be required; however, the preoperative diagnosis of these growths is difficult. Here, we report a case of cavernous hemangioma of the small intestine that was diagnosed using wireless capsule endoscopy and single-balloon enteroscopy. Our patient presented with melena and iron deficiency anemia. Neither gastroscopy nor colonoscopy detected any remarkable findings. Thus, we performed wireless capsule endoscopy and single-balloon enteroscopy, which revealed a blue submucosal lesion (length, 2 cm) with a small red spot on its surface in the distal jejunum. Accordingly, we diagnosed the lesion as a cavernous hemangioma. Laparoscopic-assisted small intestinal resection was performed successfully. This case highlights the usefulness of wireless capsule endoscopy of the small intestine as a diagnostic tool for preoperative detection of the causes of obscure gastrointestinal bleeding, including cavernous hemangioma of the small intestine.

Case Report

A 56-year-old woman visited her local hospital because of worsening fatigue and melena that had persisted for a week. Gastroscopy and colonoscopy were performed but no active bleeding or lesions were detected. Because the patient’s symptoms persisted, she was referred and admitted to our hospital for further evaluation 50 days after her first visit to her local hospital. The woman had a history of surgery for uterine fibroid tumors and appendicitis. Furthermore, she had been receiving treatment for articular rheumatism at her local hospital. She had not been taking nonsteroidal anti-inflammatory or antiplatelet drugs.

On admission, physical examination of the patient revealed pale conjunctivae. A clinical examination of the abdomen did not detect any pain, masses, or vascular bruits. However, laboratory analysis revealed marked anemia (hemoglobin, 6.7 g/dL), and low serum iron and ferritin levels. The patient received a blood transfusion, and her hemoglobin levels improved (> 10 g/dL). An investigation of her small intestine was performed using a wireless capsule endoscope, and an elevated red lesion was found in the jejenum (Fig. 1a). The lesion was not bleeding, and no other lesions were detected in the small intestine. On contrast-enhanced computed tomography, the mass (diameter, 2 cm) showed enhancement and was located in the pelvic region of the small intestine (Fig. 1b). Single-balloon enteroscopy performed using an antegrade approach revealed that the lesion was located in the distal jejunum, 1.5 m from Treitz’s ligament. It was approximately 2 cm in diameter, appeared to be a submucosal tumor, and was blue with superficial red spots (Fig. 2). Based on the findings from wireless capsule endoscopy and single-balloon enteroscopy, we diagnosed the lesion as a cavernous hemangioma.
On the 20th hospital day, laparoscopy-assisted small intestinal resection was performed with jejuno-jejunal reanastomosis. Macroscopic examination of the resected specimen revealed that the lesion measured 1.3 × 1.0 cm and was elastic, soft, and purplish-blue. Pathological examination showed vascular proliferation within the submucosa; that is, large, dilated, blood-filled vessels lined by flattened endothelia (some of which displayed thrombotic phenomena) were observed (Fig. 3). The histological diagnosis was cavernous hemangioma of the small intestine. Postoperatively, the patient recovered well and her symptoms have not recurred.

**Discussion**

Cavernous hemangioma of the small intestine is a rare disease, accounting for 5% to 10% of all small-bowel benign neoplasms [3]. Hemangioma accounted for 19 cases of 676 small intestinal tumors reported between January 1995 and December 1999 [4]. Of 144 cases of small intestinal tumors detected with double-balloon endoscopy between September 2000 and December 2005, hemangiomas were identified in 3 cases [5]. Although it is an uncommon cause of gastrointestinal bleeding, hemangioma of the small bowel often leads to the development of acute hemorrhage [6] or chronic anemia [7]. We retrieved from the PUBMED and i-chu-shi (Japan) databases reports of cavernous hemangiomas presenting with gastrointestinal bleeding that were published beginning in 2000; 46 cases (22 women, 24 men; mean age, 34.6 years) were retrieved and reviewed. The most common site of small intestinal hemangiomas was the jejunum (46%), and melena was observed in 65% of the cases. The mean diameter of the lesions was 2.93 cm. Twenty-two of the 46 lesions (48%) were diagnosed preoperatively (Table 1) [2, 3, 7, 8]. Of these cases, seven were detected with capsule endoscopy and 10 were diagnosed using balloon enteroscopy. Compared with the cases reported before 2000, a markedly increased proportion of cases were diagnosed preoperatively using capsule endoscopy and balloon enteroscopy from 2000 onward. According to the algorithms for the diagnosis and treatment of obscure gastrointestinal bleeding proposed by the American Gas-
Competing interests: None

Table 1  Reports since 2000 on preoperative diagnosis of small intestinal cavernous hemangiomas.

| Author, Year | Age (years) | Sex | Symptom | Preoperative examination | Hemangioma size (cm) | Hemangioma location | Treatment |
|--------------|-------------|-----|---------|--------------------------|-----------------------|---------------------|-----------|
| Shimizu et al., 2006 | 44 | M | Melena | Computed tomography | 2.0 | Ileum | Laparotomy |
| Fukumura et al., 2006 | 9 | F | Anemia | Colon endoscopy | 1.0 | Multiple | Laparotomy |
| Zeng et al., 2008 | 21 | M | Abdominal pain | Computed tomography | 2.2 | Jejunum | Laparotomy |
| Deng et al., 2008 | 6 | M | Melena | Balloon enteroscopy | 1.0 | Multiple | Conservative treatment |
| Deng et al., 2008 | 6 | M | Balloon enteroscopy | 1.0 | Multiple | Conservative treatment |
| Deng et al., 2008 | 7 | M | Melena | Balloon enteroscopy | 1.0 | Multiple | Conservative treatment |
| Willert et al., 2008 [3] | 19 | M | Anemia | Capsule endoscopy + balloon enteroscopy | 1.4 | Multiple | Endoscopic treatment |
| Pinho et al., 2009 [2] | 9 | F | Anemia | Capsule endoscopy | 2.5 | Ileum | NA |
| Tsutsui et al., 2009 | 40 | F | Anemia | Balloon enteroscopy | 5.0 | Jejunum | Laparotomy |
| Morita et al., 2009 | 69 | M | Anemia | Capsule endoscopy + balloon enteroscopy | 0.5 | Ileum | Laparoscopic operation |
| Sako et al., 2009 | 40 | F | Melena | Computed tomography | 6.0 | Jejunum | NA |
| Endo et al., 2009 | 49 | F | Melena | Small intestinal imaging | 1.2 | Multiple | Endoscopic treatment |
| Takayama et al., 2010 | 71 | F | Abdominal pain | Computed tomography | 3.0 | Ileum | Laparotomy |
| Abdul Aziz et al., 2011 | 6 | F | Abdominal pain | Ultrasonography | 15 | Ileum | Laparotomy |
| Rodrigues-Zentner et al., 2011 | 46 | M | Anemia | Colon endoscopy | 2.3 | Ileum | Laparoscopic operation |
| Mikami et al., 2011 | 45 | F | Melena | Capsule endoscopy + balloon enteroscopy | 0.9 | Multiple | Endoscopic treatment |
| Pera et al., 2012 [7] | 16 | M | Anemia | Capsule endoscopy + balloon enteroscopy | 4.2 | Jejunum | Laparoscopic operation |
| Guardiola et al., 2012 | 19 | M | Melena | Capsule endoscopy | 1.0 | Ileum | Laparoscopic operation |
| Miyamoto et al., 2012 | 61 | F | Anemia | Computed tomography | 4.0 | Ileum | Laparotomy |
| Dhumane et al., 2013 [8] | 60 | M | Anemia | Capsule endoscopy + balloon enteroscopy | 7.0 | Jejunum | Laparoscopic operation |
| Tanioka T et al., 2013 | 16 | F | Anemia | Balloon enteroscopy | 1.5 | Jejunum | Laparoscopic operation |
| Sato M et al., 2013 | 9 | F | Abdominal pain | Computed tomography | 2.5 | Jejunum | Laparotomy |
| Our case, 2013 | 56 | F | Melena | Capsule endoscopy + balloon enteroscopy | 1.3 | Jejunum | Laparoscopic operation |

M, male; F, female; NA, not available

Endoenterological Association in 2007, capsule endoscopy should be used for the initial examination. When positive findings are acquired, balloon enteroscopy should be performed [9]. In the current case, capsule endoscopy and balloon enteroscopy were performed based on these guidelines, and we were able to detect the characteristic findings of cavernous hemangioma, e.g., a blue submucosal lesion with a bleeding spot on its surface.

Regarding the treatment of bleeding hemangiomas, most of the previous cases were treated surgically [10]. Endoscopic treatment was performed in only three cases. One of the hemangiomas was clipped, another was subjected to sclerotherapy, and the third was removed by means of snare polypectomy [3]. In these three cases, bleeding occurred frequently, and the multiple lesions were relatively small. In our case, we did not perform endoscopic treatment because active bleeding was not present at that time, and there seemed to be a risk for massive bleeding after endoscopic treatment. Future studies are needed to determine the indications for endoscopic treatment.

In conclusion, we encountered a case of cavernous hemangioma of the small intestine that was diagnosed preoperatively using wireless capsule endoscopy. Capsule endoscopy is clearly useful for preoperative diagnosis of hemangiomas in the small intestine.