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Cavernous hemangioma of the ileum ² in a young man: A case report and review of literature

Li Y *et al.* Cavernous hemangioma of the ileum

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Abstract

BACKGROUND

Small intestinal cavernous hemangioma is a rare disease, especially in the ileum. It is difficult to accurately diagnose due to its hidden location and non-specific clinical symptoms. Herein, we reported a case of ileal cavernous hemangioma with chronic hemorrhage in a 20-year-old man and reviewed the literature to gain a better understanding of this disease.

CASE SUMMARY

The patient complained of intermittent melena and hematochezia for more than three months. The lowest hemoglobin level revealed by laboratory testing was 3.4 g/dL (normal range: 12-16 g/dL). However, the gastroscopy, colonoscopy, and peroral double-balloon enteroscopy (DBE) showed no signs of bleeding. The trans-anal DBE detected a lesion at about 340 cm proximal to the ileocecal valve. Thus, we performed an exploratory laparoscopy and the lesion was resected. After the operation, the patient had no melena. Finally, the pathological examination identified the neoplasm as an ileal cavernous hemangioma, thereby resulting in gastrointestinal hemorrhage.

CONCLUSION

This report might improve the diagnosis and treatment of ileal cavernous hemangioma.

Key Words: Cavernous hemangioma; Ileum; Gastrointestinal hemorrhage; Anemia; Double-balloon enteroscopy; Contrast-enhanced computed tomography; Case report

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Core Tip: Small intestinal hemangiomas are rare benign tumors, and ileal cavernous hemangiomas are even rarer. Here, we reported a 20-year-old male with intermittent melena and hematochezia for more than three months. We successively performed a colonoscopy, gastroscopy, contrast-enhanced computed tomography, trans-oral double-balloon endoscopy, and trans-anal double-balloon endoscopy, and finally found a lesion in the ileum. The lesion was resected by laparoscopy. Thus, to further improve the diagnosis and treatment of this disease, we collected cases of ileal cavernous hemangioma in the past 20 years and summarized its manifestations, complications, imaging features, endoscopic features, and therapeutic strategies.

INTRODUCTION

Small intestinal hemangiomas are rare vascular malformations and account for 7 to 10% of all benign tumors in the small intestine. Among them, 60% occur in the jejunum, being rare in the ileum^[1,2]. Histologically, small intestinal hemangioma is a congenital vascular malformation¹ that can be classified as capillary, cavernous, or mixed-type, with cavernous hemangioma being the most prevalent^[3]. It can be manifested as abdominal pain, gastrointestinal bleeding, intestinal obstruction, intussusceptions, and intestinal perforation in some patients^[4,5,6]. Due to the lack of specific clinical symptoms and distinctive auxiliary examination characteristics, omission or misdiagnosis might occur. Recently, with the advancements in enteroscopy, the detection rates of small bowel diseases have increased, but rare cases of small intestinal cavernous hemangioma, especially cavernous hemangioma of the ileum, are still misdiagnosed. Herein, we reported a case of gastrointestinal bleeding caused by ileal cavernous hemangioma and summarized its manifestations, complications, imaging features, endoscopic features, and therapeutic strategies in the past 20 years.

CASE PRESENTATION

Chief complaints

On June 11, 2021, a 20-year-old male was hospitalized for intermittent melena and hematochezia that had persisted for more than three months.

History of present illness

For three months before hospitalization, the patient experienced intermittent melena between 50-200 g, up to 2 times per day, accompanied by dizziness and fatigue. He was admitted to the department of hematology of our hospital, and the laboratory testing revealed severe anemia (hemoglobin, 5.7 g/dL), with microcytic and hypochromic anemia. Thus, iron deficiency anemia was diagnosed. The treatment with blood transfusion and ferrous gluconate syrup improved the anemia-related symptoms and the hemoglobin levels increased to 6.0 g/dL. For diagnosis and treatment, he was admitted to our department two months ago. His hemoglobin level was 6.2 g/dL, and the fecal occult blood test was positive. The gastroscopy, colonoscopy, and contrast-enhanced computed tomography (CECT) examination suggested no specific abnormalities. Hence, a small bowel endoscopy examination was recommended to determine the cause of bleeding, but the patient refused and was discharged. Nonetheless, the melena relapsed 9 days ago, averaging about 300 g each time, accompanied by dizziness and aggravated fatigue. He was sent to a nearby hospital and had a hemoglobin level of only 3.4 g/dL. After a blood transfusion, the symptoms of anemia were slightly improved. Four days ago, the melena relapsed again, and the situation appeared to have significantly deteriorated and he returned to the department of gastroenterology of our hospital.

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History of past illness

The patient had no past medical history.

Personal and family history

The patient did not present previous personal and family medical history.

Physical examination

At admission, his heart rate was 92 beats per min and his blood pressure was 12.1/6.9 kPa. He was pale, and the other physical examinations were normal.

Laboratory examinations

The laboratory testing ¹revealed moderated microcytic and hypochromic anemia (hemoglobin: 6.95 g/dL). The fecal occult blood test was positive. The liver ⁴and kidney function, coagulation function, carcinoembryonic antigen (CEA), carbohydrate antigen 19-9 (CA19-9), and cancer antigen 125 (CA125) were normal.

Imaging examinations

Endoscopy re-examination: the gastroscopy showed that the corpus had a spot of flushing and erosion, but there was no bleeding, and the stomach cavity was clean. The colonoscopy revealed dark red blood at the end of the ileum. After flushing with normal saline, there were no abnormalities in the mucosa, but dark red blood continued to gradually drip down from the ileum, indicating small intestinal bleeding. The CECT re-examination (SOMATOM Definition Flash, Siemens, Germany) showed a lesion in the ileum that was enhanced in the arterial and venous phases (Figure 1A-D).

Peroral double-balloon enteroscopy (DBE): mild inflammation of the upper jejunum was detected. The trans-anal DBE reached the upper segment of the ileum. In the middle and lower segments, the mucosa was rough and eroded, and the villi were absent. A submucosal lesion with approximately 1.2 cm x 1.0 cm was found at about 340 cm proximal to the ileocecal valve with ulcer formation (Figure 1E&F). The surrounding mucosa was flushed, presented edema, was hard in texture, easy to bleed, and had numerous thickened blood vessels supplying the tumor. The endoscopic diagnosis was: 1. Ileal submucosal bulge (Stromal tumor? Vascular malformation?) 2. Ileal inflammation.

FINAL DIAGNOSIS

Our clinical diagnosis was small intestinal vascular malformation. The postoperative pathological diagnosis revealed ileal cavernous hemangioma.

TREATMENT

First, a laparoscopic exploration was performed and a tortuous collection of irregular vessels was found at about 300 cm proximal to the ileocecal valve. Then, the intestine was longitudinally opened near the irregular vessels. A 1.3 cm x 1.2 cm submucosal lesion with an ulcer (0.3 cm x 0.5 cm) and pulsatile bleeding on the surface was found in the ileum. The lesion was completely resected. The optical microscopy showed twisted and dilated blood vessels in the submucosa, surrounding the bleeding onset. Finally, the pathological diagnosis was cavernous hemangioma of the ileum.

1 OUTCOME AND FOLLOW-UP

The patient quickly recovered and had no further episodes of bleeding after the operation. He returned to our hospital for a follow-up visit one month after the operation and his hemoglobin levels had increased to 10.7 g/dL (normal range: 12-16 g/dL).

DISCUSSION

Small intestinal bleeding accounts for 5 to 10% of all gastrointestinal bleedings^[7]. Hence, it should be considered for persistent or recurrent bleedings that are not detected by routine endoscopy, including upper gastrointestinal endoscopy and colonoscopy. The common etiologies of small intestinal bleeding in individuals younger than 40 include Crohn's disease, tumor, Merkel's diverticulum, Dieulafoy lesions, vasodilatory disorder, and polyp syndrome^[7].

Small intestinal hemangiomas are rare vascular malformations that can cause small intestinal hemorrhages. They can occur in various segments of the small intestine, but are more frequent in the jejunum and rare in the ileum^[8,9,10]. Pathologically, hemangiomas are classified into capillary, cavernous, and mixed types, with capillary types being the most common^[3]. Here, we presented a case of cavernous hemangioma in the upper ileum causing recurrent gastrointestinal bleeding.

Furthermore, a small intestinal hemangioma can originate from the mucosal, submucosa, muscularis propria, or even the serosal layer^[11]. Its most common manifestations include recurrent gastrointestinal bleeding and iron deficiency anemia, which might be accompanied by non-specific symptoms such as dizziness and fatigue^[12]. Generally, no specific findings are observed in the laboratory testing, making it difficult to diagnose. Thus, to analyze the clinicopathological features of ileal cavernous hemangiomas, we searched PubMed (<https://pubmed.ncbi.nlm.nih.gov/>) and Google Scholar (<https://scholar.google.com/>) databases between 2001 and 2021 using “hemangioma”, “ileum”, “intestinal”, and “cavernous” as keywords. The language was limited to English. Case reports were included if the final diagnosis was ileal cavernous hemangioma. The exclusion criteria were: non-English literature; the location of the hemangioma was not mentioned; non-human ileal cavernous hemangioma.

A total of 15 cases (9 women and 6 ¹ men; mean age: 30.3 years) were retrieved and reviewed (Table 1). The most common symptoms (73.3%) were related to anemia, including fatigue, weakness, pallor, dizziness, loss of consciousness, and even syncope. Melena was the second most common symptom (40.0%). Non-specific symptoms of the digestive system (33.3%) included abdominal pain, nausea, vomiting, bloating, abdominal mass, and loss of appetite (Table 1). Among cases, a 6-year-old girl developed hemangioma hemorrhage after a mild abdomen blunt trauma, and a diagnostic cum therapeutic laparoscopy was performed^[14]. Our current patient had melena as the main symptom, accompanied by anemia-related dizziness and fatigue, consistent with the literature.

Nine of the 15 Lesions (60%) were diagnosed by endoscopy before operations. Among them, five were detected by capsule endoscopy (CE), two by DBE, one by both CE and DBE, and one in the terminal ileum by colonoscopy. Interestingly, in our current case, the colonoscopy revealed dark red blood near the end of the ileum and the fecal occult blood was positive. However, Dália Fernandes *et al* performed a colonoscopy and ileoscopy during asymptomatic disease and discovered no sign of blood or bleeding lesions^[15]. Therefore, we hypothesized that conducting an emergency endoscopy during the active stage of the disease might improve diagnostic performance. Regarding the endoscopic characteristics of ileal cavernous hemangioma in the literature reviewed, five cases were represented as submucosal lesions, four as polypoids, two as irregular nodules that can be easily misdiagnosed as cancers, and one as a big heterogeneous mixed lesion. Three case reports did not describe the morphology of lesions in detail. The surface of the lesion can present erythema, thrombosis, or bleeding. The color of the lesion was mostly reddish-purple, blue, and purple-blue, indicating a vascular network. The patient reported here was also represented by a submucosal lesion, but with ulcers on the surface and no blue-purple appearance, thrombosis, or bleeding, and could be easily misdiagnosed as a stromal tumor, especially a malignant stromal tumor. Therefore, we should strengthen our understanding of the characteristics of ileal cavernous hemangioma to prevent misdiagnoses. Computed tomography (CT) is a non-invasive examination but has limited significance in diagnosing small intestinal hemangioma. On the other hand, CT can be helpful to initially determine the location and general shape of the lesion and guide the next step of endoscopy or surgery. Thus, we summarized the imaging features of these fifteen patients: 33.3% (5/15) presented intestinal wall thickening and 20.0% (3/15) presented calcifications. These results were different from Ji-xin Fu *et al*, in which 50% of patients had phleboliths^[12]. Herein, the CT imaging of our patient presented wall thickening and enhancement in the arterial and venous phases, consistent with previous reports^[27]. Then, we investigated the hemangioma size of five patients with wall thickening of the ileum (one case did not mention the size^[15]). The

median hemangioma length of these patients was 4 cm, and the mean diameter was 9.5 cm. Besides, the lesions either originated from the submucosa or reached the submucosa. Thus, we suggest that the thickening of the intestinal wall might indicate the size and depth of the lesions. Furthermore, among the cases reviewed, one had secondary intussusception with perforation^[15], consistent with previous reports^[4].

In these fifteen cases, various preoperative diagnostic methods were used, including CECT (46.7%), 99m-Tc-RBC-scintigraphy (20%), and magnetic resonance imaging (MRI) (6.7%). The CECT can clearly show the structures of the intestinal wall and extra-intestinal lesions in multiple directions and angles, which can initially be used to determine the location and general shape of the lesions, contributing to more information for clinical diagnosis. Moreover, CE and DBE are used for diagnosing small bowel diseases and there is no significant difference in the overall diagnosis rate between these methods (50 and 53%, respectively)^[28]. CE is non-invasive, painless, and tolerable for the patient^[29]. However, disadvantages might also exist due to the inability of tissue biopsy or endoscopic treatment, inaccurate positioning, and being unsuitable for patients with massive bleeding or intestinal obstruction^[30,31]. Meanwhile, DBE has intuitiveness, operability, biopsy feasibility, as well as the efficacy of endoscopic treatment, which significantly improves the diagnosis and treatment of small bowel diseases. However, the high cost, invasiveness, and risk of acute pancreatitis or intestinal perforation in DBE should not be ignored^[32,33]. Endoscopic treatments such as electrocoagulation and titanium clamp hemostasis are suitable for some small intestinal bleeding caused by vascular diseases. For hemangioma, endoscopy biopsy is not recommended, since it can easily lead to uncontrollable bleeding. In our current case, the patient presented active bleeding, and the cause of gastrointestinal bleeding was highly suspected as vascular malformations. Thus, we selected DBE to diagnose and perform the endoscopic treatment.

Polidocanol injection using DBE is a safe, feasible, and effective treatment method for small intestinal hemangioma^[34]. A retrospective cohort study showed that 66.3% of individuals improved after receiving DBE polidocanol injections. Additionally, in

younger patients, the effectivity rate is 100% during long-term follow-up^[35]. However, for large or diffuse hemangiomas, or lesions originating from the submucosa or deeper layers, the endoscopic procedure might result in perforation and uncontrolled bleeding. In this case, surgical resection might be safer^[1] and was selected to finally treat our patient.

CONCLUSION

Small intestinal cavernous hemangioma is a rare disease. Its most common symptoms are anemia and melena. CECT can initially be used to determine the location and general shape of the lesions. The wall thickening and high-density shadow in the CECT might have diagnostic value for small intestinal cavernous hemangiomas. CE and DBE can contribute to the identification of lesions, mostly represented as submucosal masses, polypoid or irregular nodules. A bluish-purple vascular network is generally visible on the surface of a typical lesion, and atypical lesions are easily misdiagnosed as stromal tumors and other cancers. Regarding the detection rate, there is no significant difference between CE and DBE, but, since CE is non-invasive, it is more recommended to detect the lesions. Both endoscopy and surgery are feasible for this disorder. However, for large or diffuse hemangiomas or lesions derived from the submucosa, surgical resection might be chosen. A detailed treatment plan for each case should be prepared to effectively manage patients with variable risks. Finally, more case reports or observational studies are needed to help in the diagnosis and treatment.

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SIMILARITY INDEX

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