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CASE REPORT

Endoscopically diagnosed cavernous hemangioma in the deep small intestine: A case report

Hsin-Hung Chen [a], Chia-Hung Tu [a,*], Po-Chu Lee [b], Han-Mo Chiu [a], Ming-Shiang Wu [a], Hsiu-Po Wang [a]

[a] *Department of Internal Medicine, National Taiwan University Hospital, Taipei, Taiwan*
[b] *Department of Surgery, National Taiwan University Hospital, Taipei, Taiwan*

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Summary We report a 27-year-old female with chronic iron deficiency anemia and unexplained fecal occult blood. Abdominal ultrasonography and computed tomography disclosed a possible endoluminal lesion in the small intestine. Single-balloon enteroscopy detected the target lesion in the proximal ileum. The lesion was a 2.5-cm submucosal tumor that was purple-red, soft, had a narrow base, and exhibited superficial telangiectasia. After endoscopic marking, the tumor was resected with minimally invasive laparoscopy. It was histologically confirmed as a cavernous hemangioma. In this report, we discuss the endoscopic characteristics, surgical and pathological assessment, and management strategy of hemangiomas in the small intestine.

Introduction

Hemangioma is rare among primary small intestinal tumors and can present as an infrequent cause of obscure gastrointestinal bleeding [1,2]. A preoperative diagnosis has seldom been possible owing to the tumor's deep location that is often beyond the reach of regular endoscopy [2]. Thus, most cases reported in the literature were diagnosed with surgical exploration. With the recent advent of newer techniques such as balloon-assisted enteroscopy [3], endoscopic diagnosis or even resection of small intestinal hemangiomas is becoming feasible. However, the proposed diagnostic features vary between experienced endoscopists, and the evolution of nonsurgical

* Corresponding author. Department of Internal Medicine, National Taiwan University Hospital, Number 7, Chung-Shan South Road, Taipei 100, Taiwan.
E-mail address: 601533@ntuh.gov.tw (C.-H. Tu).

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treatment options has led to new controversy [4,5]. We report a case of a presurgically diagnosed ileal hemangioma as a source of chronic gastrointestinal blood loss, demonstrate its endoscopic features, and discuss the risk of endoscopic tumor resection from a pathological prospective.

Case report

A 27-year-old female was referred to our gastroenterology department for obscure gastrointestinal bleeding. Prior to referral, she had been continuously treated in the hematology clinic for unexplained chronic iron deficiency anemia since she was a teenager. Over the years, her treatment consisted of oral iron therapy and occasional transfusions. She experienced significant anemic symptoms episodically, which somewhat interfered with her social life. She had denied abdominal symptoms, melena, hematochezia, bowel habit changes, and body weight loss. She was on no medications, other than iron preparations. On presentation, physical examination was only remarkable for generalized pallor. Laboratory studies showed a hemoglobin level of 4.6 g/dL, a mean corpuscular volume of 60.5 fL, and decreased iron stores [iron level, 9 μg/dL (normal range, 50—212 μg/dL); total iron-binding capacity, 392 μg/dL (275—332 μg/dL); transferrin saturation, 2.29%; ferritin level, 1.99 ng/mL (3—151 ng/mL)]. Hemoglobin electrophoresis results were normal. In the previous 5 months, her transfusion requirements had increased. During this time, four separate fecal occult blood tests were positive. However, esophagogastroduodenoscopy and colonoscopy did not identify any potential source of blood loss. Therefore, a bleeding source in the small intestine was suspected.

We first performed intestine-targeted transabdominal ultrasonography, which disclosed a questionable 2-cm hypoechoic, compressible, endoluminal tumor superficially located 5 cm below the umbilicus (Fig. 1A). Subsequent computed tomography (CT) further characterized the lesion as a 2.5 cm contrast-enhancing, small intestine-bound tumor with internal calcifications in the lower peritoneal cavity (Fig. 1B). There were no other findings or additional calcifications along the intestines.

Enteroscopy was performed for presurgical marking, and possible histological evaluation. Using a SIF-Q260 enteroscope with ST-SB1 splinting tube (Olympus Co., Tokyo, Japan) via an antegrade approach, the target lesion was successfully accessed near the insertion limit of the endoscope. This location was judged as the proximal ileum (approximately 300 cm beyond ligament of Treitz) under fluoroscopic guidance (Fig. 2A). The lesion was a purple-red, soft, depressible, semipedunculated tumor that exhibited superficial telangiectasia (Fig. 2B and C). Biopsy was thought unnecessary.

Endoscopic tattoos with India ink were applied adjacent to the lesion. Additional marking with hemoclipping was applied to allow intraoperative fluoroscopy guidance whenever necessary.

Surgical resection of the tumor was performed on the day after enteroscopy, using the minimally invasive single-port laparoscopy technique [6]. The lesion was easily visualized at surgery, and appeared as a well-defined reddish bulge on the antimesenteric ileal serosa (Fig. 3A). There was no additional finding during surgical exploration. Ileal segmental resection was performed. The main bulk of the tumor was endoluminal (Fig. 3B).

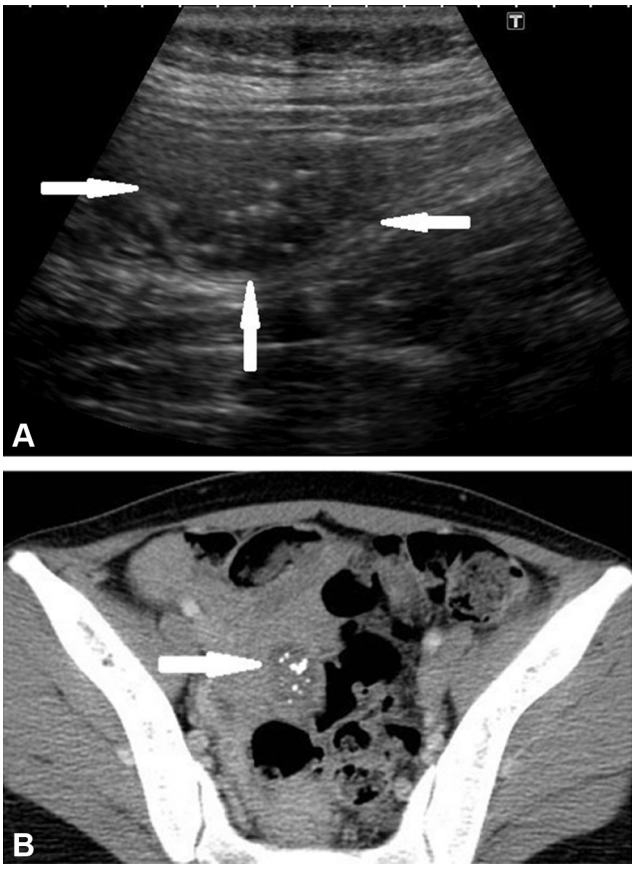

Figure 1 (A) Transabdominal ultrasonography shows a 2-cm hypoechoic lesion (arrows) within the ileal lumen, which contains internal calcifications (that appear as bright dots). (B) Computed tomography reveals that the lesion is 2.5 cm with soft tissue density and scattered internal calcifications arising from the small intestine (arrow).

Histological examination confirmed a cavernous hemangioma (Fig. 4). At the 4-month follow up, the patient exhibited a spontaneously rising hemoglobin level, which indicated successful cessation of blood loss.

Discussion

Hemangioma of the small intestine accounts for 7—10% of all small intestinal benign neoplasms [1,2,7]. It is histologically classified as "cavernous", "capillary", or "mixed-type", based on the size of the affected vessels [2]. Occult chronic gastrointestinal bleeding and iron deficiency anemia are the more common presentations, whereas intussusception and intestinal obstruction are less common presentations [2]. Technical advances have allowed