Jejunal bleeding: a case report

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Abstract

Small intestinal bleeding remains a relatively uncommon event, accounting for 5-10% of all patients presenting with gastrointestinal bleeding. A rare but significant source includes hemangioma of the small intestine. Here we present a case of a 78-year-old female patient admitted with recurrent melena and iron deficiency anemia. After performing a normal upper and lower endoscopic examination, we suspected small bowel bleeding. Correspondingly, the patient underwent a video capsule endoscopy, showing a submucosal nodular lesion in the distal jejunum. Balloon enteroscopy clearly identified and inked the lesion, facilitating minimally invasive surgery. Pathology demonstrated a cavernous hemangioma. As we did not have a conclusive diagnosis preoperative and endoscopic intervention may have led to uncontrolled bleeding or perforation, laparoscopy was chosen to conduct a better evaluation. The present case findings emphasize that gastrointestinal hemangiomas, although uncommon, should be considered in the differential diagnosis of patients who present with unexplained gastrointestinal bleeding or other abdominal symptoms. (Acta gastroenterol. belg., **2023,** 86, **567-570**.

Keywords: hemangioma, small intestine, gastro-intestinal bleeding, obscure.

Introduction

Hemangioma of the small intestine is a rare benign hereditary vascular malformation. Small bowel hemangiomas can be completely asymptomatic or cause occult or obscure gastrointestinal bleeding. The majority of patients present with occult gastrointestinal bleeding and iron deficiency anemia. Endoscopy may be very helpful for differential diagnosis and remains the primary diagnostic technique for mass-forming lesions. Upper endoscopy and colonoscopy frequently reveal these lesions, but many occur in the mid-to-distal parts of the small bowel and can be difficult to identify. In patients with recurrent melena, current guidelines recommend video capsule endoscopy as a first-line procedure for small bowel evaluation after second-look upper and lower gastro-intestinal sources have been ruled out. Balloon enteroscopy can be used when endoscopic evaluation and/or therapy is required based on similar diagnostic yields. Macroscopically, cavernous hemangiomas appear as a polypoid or mound-like reddish purple mucosal lesions. Although surgical intersection is the standard approach, endoscopic treatment has been performed successfully in many patients with hemangiomas. It is generally prudent and might be suitable for multiple, relatively small lesions. We describe a case with solitary small bowel hemangioma, which was diagnosed preoperatively by video capsule endoscopy and was clearly identified by enteroscopy, facilitating minimally invasive surgery.

Case report

A 78-year-old female patient, with a known history of osteoporosis, presented to the emergency department with dyspnea, fatigue and melena since a few days. Recently, non-vitamin K antagonist oral anticoagulant therapy was started because of atrial fibrillation. Physical examination showed a patient with pale conjunctivae with a soft and non-tender abdomen. Furthermore a normal blood pressure and oxygen saturation in association with tachycardia (117') was observed. Her family history was negative. There was no significant alcohol, tobacco or drug abuse.

Initial routine blood analyses reported a normocytic hypochromic anemia with iron deficiency: hemoglobin 4,9g/dL (normal range: 11,9-14,6g/dL), hematocrit 16,2 % (normal range: 36.6-44,0%), MCV 86,2 fL (normal range: 82,9-98,0 fL) and ferritin <5 µg/L (normal range: 15-150 µg/L). Two months before, the hemoglobin level was 11,4g/dL with a hematocrit of 35,6%. Furthermore normal folic acid and vitamin B12. There was no renal insufficiency observed.

Gastro-, duodeno-, and sigmoidoscopy were normal without any signs of bleeding. Fecal occult blood test (FOBT) was positive. A colonoscopy was planned in the nearby future. A few weeks afterwards, the hemoglobin level dropped repeatedly to 5 g/dL accompanied by recurrent melena. Gastro- and duodenoscopy were performed again and remained without abnormalities. Colonoscopy showed a small caecal adenoma without signs of bleeding. Therefore a polypectomy was performed. Through the ileal valve, a large amount of blood was seen. Video capsule endoscopy was performed since there was no evidence of obstruction. It showed an active bleeding in the jejunum. So antegrade enteroscopy was planned and confirmed a submucosal nodular lesion in the distal jejunum (figure 1a). The lesion was marked with lipiodol. No additional lesions were found. CT

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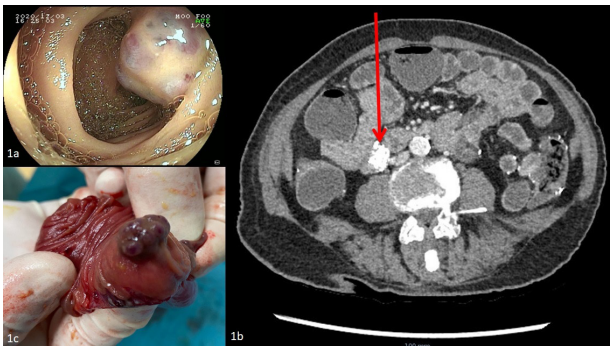

Figure 1. — a: Antegrade enteroscopy showed a submucosal nodular lesion in the distal jejunum. b: Pre-operative computed tomography (CT) scan: Hemangiomas (red arrow) are found to be hypervascular with a peripheral, progressive and discontinuous contrast uptake in the arterial phase on CT. c: Macroscopic appearance of the resected specimen. The specimen was bluish-purple in colour, with cystic nodules of varying sizes fused into a tumour.

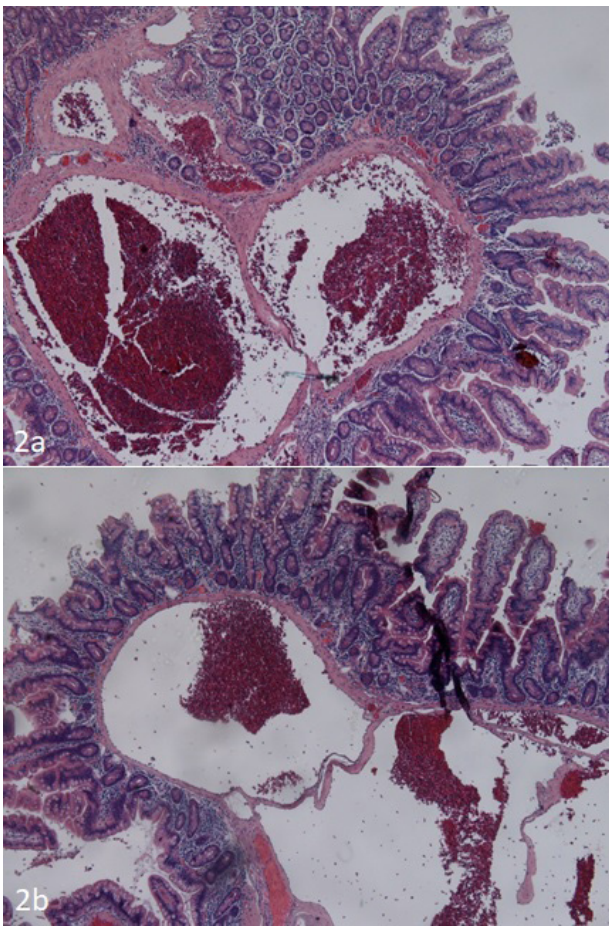

Figure 2. — a-b : Microscopic examination of haematoxylin and eosin-stained resected tumour tissue sections revealed lumens of different sizes, shapes and thicknesses, and the presence of disintegrated red blood cells.

enterography showed an intraluminal hyperdense structure in the intestines with a diameter of 2,4cm. No adenopathies and no extrinsic or mesenterial extension were seen (figure 1b). Considering the lesion was large, a biopsy, endoscopic mucosal resection (EMR) or endoscopic sclerotherapy might lead to uncontrolled bleeding, so we could not verify the diagnosis preoperatively. Therefore, laparoscopic surgery was deemed the best choice. Three days later, a laparoscopic assisted partial resection of the small intestine was performed. A bluish-purple-colored raspberry-like lesion with cystic nodules of varying sizes, fused into a tumor, was found. The lesions was completely resected (figure 1c). No other lesions were detected intraoperatively in the remaining gastrointestinal tract. Anatomopathological analysis showed a submucosal vascular lesion with dilated thin-walled vessels without signs of malignancy, confirming a cavernous hemangioma of 1,5cm diameter, which was completely removed (figure 2 a-b). The perioperative period was safe and the clinical outcome was extremely favorable after 2 years of follow-up, with a hemoglobin level of 11,9g/dL. There were no further episodes of bleeding since the operation.

Discussion

Hemangioma of the small intestine is a rare benign hereditary vascular malformation (1-7). It accounts for 5-10% of all benign tumors of the small intestine and for 0,05% of all gastrointestinal neoplasms (1,3,5,7,9). Hemangiomas are usually present at birth and have a strong tendency to develop after a period of growth in the first years of life (4). They develop from rapidly growing embryonic mesodermal tissue and have a proliferation of endothelial cells that manifest in various forms, locations and dimensions (3). Hemangiomas can arise anywhere in the luminal gastrointestinal tract. They can be localized or dispersed, the jejunum being the most common site of involvement (60,9%) (1,3,5). Most cases are single (75,7%) and small, ranging from a few millimeters to 2 cm, but larger lesions occur, especially in the rectum (8). Considered by some to be true neoplasms, hemangiomas generally are thought to be hamartomas (4,8). Intestinal hemangiomas are distinguished by slow involution and absence of recurrence (2). They are structurally complex lesions characterized by an overabundance of blood vessels, typically veins and capillaries, in a focal area of submucosal connective tissue (2,8). Hemangiomas can be classified histologically as capillary, cavernous or mixed-type based on the size of the vascular channels (1-3,8). Cavernous hemangiomas, as in our case, are most commonly found in the upper and lower gastrointestinal tract, but they can also occur in the small intestine (4). Etiologically, they originate from the submucosal vascular plexuses and can spread into the muscular layer or beyond (1). Numerous dilated, irregular blood-filled spaces can be seen microscopically within the mucosa and submucosa and they can sometimes extend through the muscular wall to the serosal surface (2,5,6,8). Macroscopically, cavernous hemangiomas appear as a polypoid or mound-like reddish purple mucosal lesion

(2,5,6,8). It is possible to visualize the mucosal oedema, nodules and vascular congestion (2). They appear morphologically as submucosal tumor-like, diffusely infiltrating or polypoid lesions (2,8). The size can vary from a few millimeters to several centimeters in the giant type (5). Cavernous hemangiomas are soft and compressible (2).

Small bowel hemangiomas can be completely asymptomatic or cause occult or obscure gastrointestinal bleeding (10). They mostly present with occult gastro-intestinal bleeding and iron deficiency anemia (1-3, 5-7,9). In our case, the recent start of anticoagulation therapy will likely have provoked the hemorrhage. Small bowel bleeding is relatively rare, comprising only about 5% of gastrointestinal bleeding (6,7). Less commonly, hemangiomas can cause complications such as intussusception, obstruction or intestinal perforation (3). Patients may also present with icterus if unusually located, such as near the ampulla of Vater (2).

Gastrointestinal hemangiomas are difficult to diagnose preoperatively, especially for small intestinal hemangiomas (7). Even small lesions can sometimes cause significant blood loss yet be very difficult to identify (10). In our case, colonoscopy suggested small bowel bleeding and video capsule endoscopy confirmed it. Endoscopy may be very helpful for differential diagnosis, and remains the primary diagnostic technique for mass-forming lesions (2,11). On CT, hemangiomas are hypervascular with a peripheral, progressive and discontinuous contrast uptake in the arterial phase (2,7). The uptake tends to be homogeneous in the portal phase (2,7). As contrast fades in the delayed phase, low-fissure or low-cobble density can be seen in the central area (2,7). The guidelines of the American College of Gastroenterology and the Korean guidelines recommend video capsule endoscopy as a first-line procedure because detection rates of video capsule endoscopy for the origins of obscure gastrointestinal bleeding are superior to CT (12,13). Because of the recent expansion of double balloon enteroscopy and video capsule endoscopy, we are able to diagnose more asymptomatic hemangiomas but also the preoperative diagnosis markedly improves (1,6,14). Although balloon-assisted enteroscopy is used to diagnose and treat many small intestinal lesions, video capsule endoscopy is now the gold standard for their diagnosis and for evaluating patients with obscure and occult gastro-intestinal bleeding, because it is noninvasive, easy to perform, and allows inspection of the entire small intestine (8,12). The inability of capsule endoscopy to perform biopsy or therapeutic intervention is its primary limitation (9).

Surgical resection is the conventional treatment modality for intestinal hemangiomas, especially for solitary lesions (1,4,5,7,14). Even large hemangiomas are compressible, facilitating minimally invasive resection (14). As in our case, applying endoscopic India ink marking prior to laparoscopic surgical resection is a particularly useful technique for more minimally invasive treatment (6). Since hemangiomas never metastasize to the lymph nodes or distant organs, local resection is sufficient (7).

Although surgery is still the primary treatment option, endoscopic treatment has been successfully in many patients with hemangiomas (9). It is generally prudent and might be suitable for multiple, relatively small lesions (1,2,4,14). Small hemangiomas, that are solitary or few in number and can be approached endoscopically. They can be coagulated using a heater probe, laser, argon plasma or bipolar electrocoagulation (2,4,8,9). Endoscopic clips may be applied to some lesions (9). Unless it is proven that the lesion is not transmural, endoscopic ablation of large lesions should be avoided (8). Local measures to control massive bleeding from cavernous hemangiomas usually are only temporarily (8). Although embolisation and surgical ligation of major feeding vessels have been used, excision frequently is required (8). Because the lesion in our case was solitary and large, laparoscopy seemed the best option. But as endoscopic therapeutic interventions improve, less invasive procedures are becoming possible (1). Of the 37 cases of intestinal hemangiomas published between 2000 and 2018, 17 cases (45,9%) were treated endoscopically. Among them, 3 cases were removed by endoscopic mucosal resection, 1 case was treated by argon plasma coagulation, and 13 cases were subjected to sclerotherapy. Most of these lesions were multiple (82,4%) and relatively small. Because intestinal hemangiomas arise from the submucosal layer and some are transmural, endoscopic treatment is risky due to the possibility of uncontrolled bleeding or perforation (1,7). Thus, endoscopic treatment of intestinal hemangiomas should be done prudently. It is important to carefully consider the indications for endoscopic treatment. A laparoscopic excision may be a better option for the large and diffuse lesions (1). Gastrointestinal hemangiomas typically have a favorable prognosis and there is no evidence on the recurrence of hemangiomas in the literature (7).

To conclude, this case report suggests to consider small bowel hemangiomas in patients presenting with unexplained gastrointestinal bleeding. Based on the literature and guideline review, we recommend that capsule endoscopy, followed by therapeutic balloon enteroscopy, be considered in patients with multiple, relatively small lesions. For the large and diffuse lesions, a laparoscopic excision might be a better approach, as in our case.

Conflict of interest

The authors have no conflicts of interest to declare.

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