

A Single Small Bowel Hemangioma Detected by Video Capsule Endoscopy in a Patient Presenting with Iron-deficiency Anemia – Two Case Reports

**Dora Grgić¹, Radovan Prijčić¹, Ivan Romić², Goran Augustin²,
Pave Markoš¹, Lea Korša³, Zlatko Marušić³, Nadan Rustemović¹,
Željko Krznarić¹**

¹Division of Gastroenterology and Hepatology, Department of Internal Medicine, University Hospital Centre Zagreb, Zagreb, Croatia;

²Department of Surgery, University Hospital Centre Zagreb, Zagreb, Croatia;

³Department of Pathology, University Hospital Centre Zagreb, Zagreb, Croatia

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Abstract: Hemangioma of the small intestine is a rare vascular malformation which mostly presents as occult gastrointestinal bleeding and iron-deficiency anemia. Patients are often asymptomatic except of fatigue due to anemia. Hemangiomas can arise anywhere in the luminal gastrointestinal tract, with jejunum as the most commonly involved site. They are very hard to recognize mostly due to their localization. Video capsule endoscopy and balloon-assisted enteroscopy have very much improved preoperative diagnostics and made major contribution to establishing the diagnosis – which was very difficult in the past and almost all cases were diagnosed during or after the operation. Surgical resection is still the conventional treatment modality, although with the improvement of endoscopic therapeutic interventions (endoscopic mucosal resection, argon-plasma coagulation) there are more therapeutic possibilities.

Mailing Address: Dora Grgić, MD., Division of Gastroenterology and Hepatology, Department of Internal Medicine, University Hospital Centre Zagreb, Kispaticeva 12, Zagreb, Croatia; e-mail: dora.grgic1@gmail.com

Introduction

We present two rare cases of an adult male patients presenting with iron-deficiency anemia for longer period of time and in whom after extensive work-up single small bowel hemangioma was found on video capsule endoscopy.

Case report 1

We present a case of a 73-years-old male patient with evident iron-deficiency anemia, which was known for several years. Patient did not have black stools, he had no symptoms of fatigue, and no hematemesis, hematochezia, abdominal pain or fever. He had not lost any weight through that period of time. He was cardiac patient (PCI LAD years before) with known arterial hypertension, with antihypertensive therapy, anti-aggregation therapy and proton pump inhibitor for his chronic medical therapy. Also, he was taking iron oral supplements during past few years. In his family his father was diagnosed with gastric cancer. Our patient had several gastroscopies done in past few years – except *Helicobacter pylori* positive gastritis that was eradicated he had no other pathological findings.

Through day hospital comprehensive gastrointestinal workup was done: laboratory findings showed mild sideropenic, microcytic anemia, and results were negative upper and lower endoscopy and unsuspecting MR (magnetic resonance) enterography. Abdominal MSCT (multi-slice computed tomography) showed hypervascularization in one of the small bowel loops and tumour was suspected. The patient was referred for small-bowel video capsule endoscopy.

Small-bowel video capsule endoscopy revealed bluish mucosal discoloration in terminal ileum which was very unusual. We performed retrograde single-balloon enteroscopy and found suspicious bluish vascular tumour-like structure 60 cm

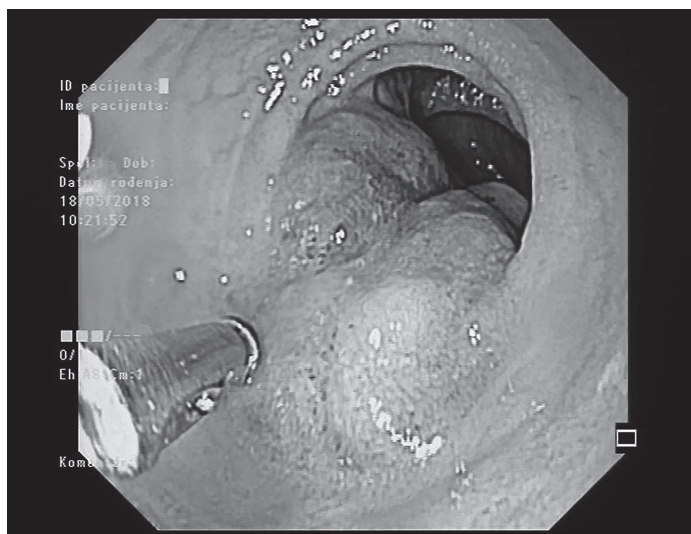


Figure 1 – Retrograde enteroscopy – tumour before taking biopsies.



Figure 2 – Loop of small intestine with tumour.

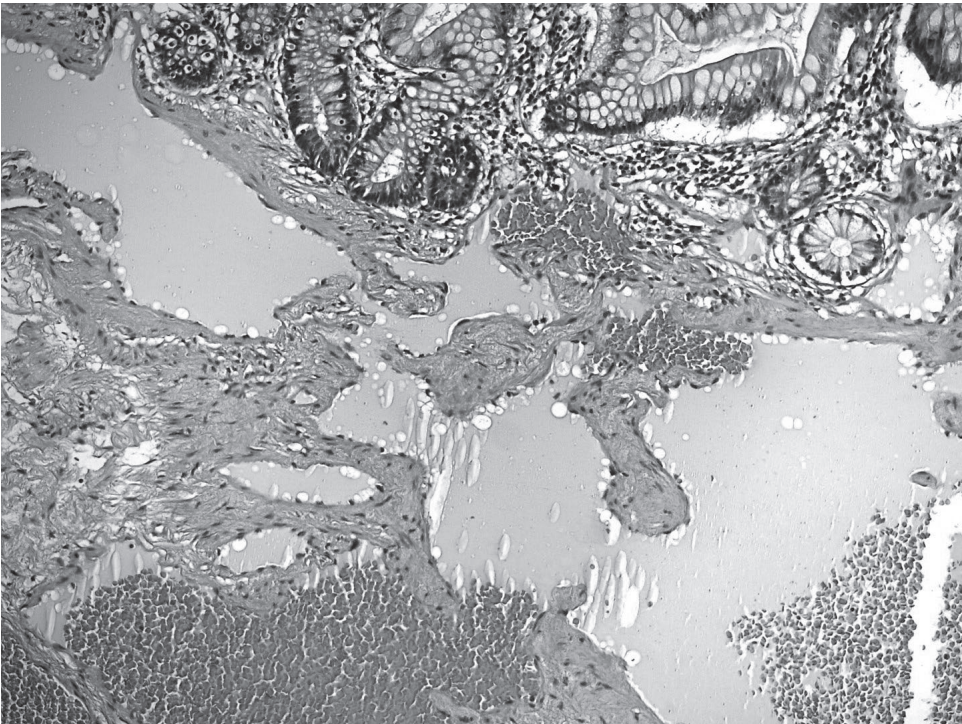


Figure 3 – Dilated tumoural blood vessels filled with erythrocytes, lined by a layer of bland endothelial cells (hematoxylin and eosin, 100×).

proximal of ileocecal (Bauchini) valve (Figure 1). Pathohistological specimens were negative for tumour cells and exploratory laparoscopy was scheduled.

During the surgery a single ileal tumour was resected (Figure 2) and histologic report confirmed intestinal hemangioma 2×2 cm in size (Figure 3).

After surgery patient recovered quickly and his haemoglobin levels in time of months increased to normal and remained stable during first year of follow-up.

Case report 2

Male patient, 63-years-old, was admitted to hospital because of microcytic anemia which was recognized several months ago. He is only taking antihypertensive therapy, and his family history is negative for gastrointestinal or other tumours.

His main symptoms were constant fatigue and weakness in everyday normal life. His colonoscopy and gastroscopy were normal, without any pathology, and because of sideropenia he received parenteral iron in 2 occasions. Because of positive test for occult bleeding video capsule endoscopy was performed and it revealed polypoid bleeding lesion in distal third of jejunum and oral third of ileum. As anatomically this area of small intestine cannot be reached with enteroscope patient was scheduled

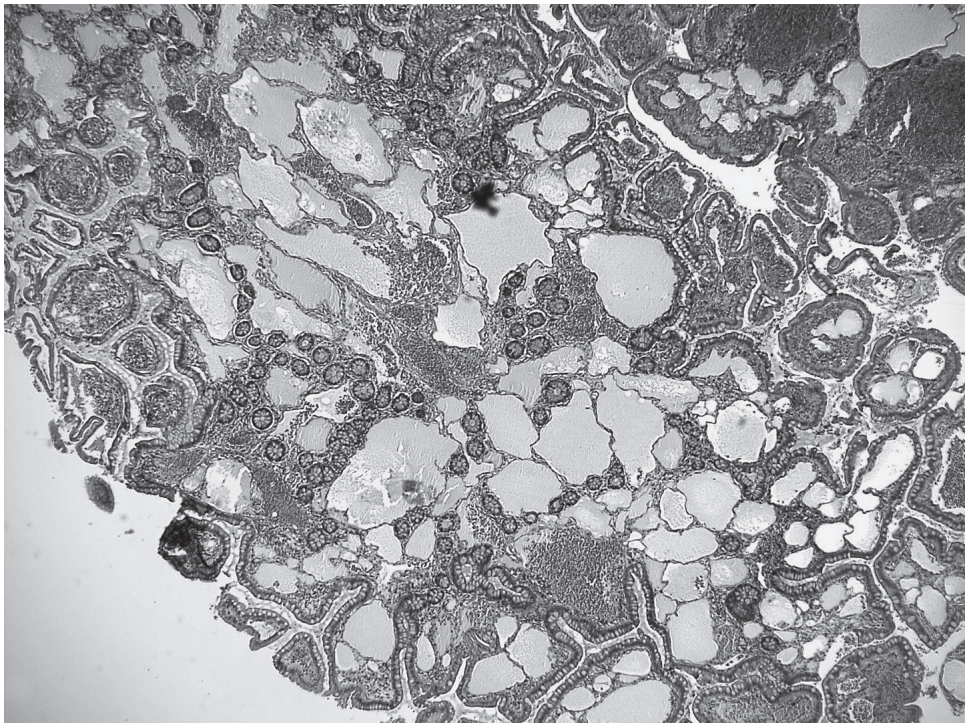


Figure 4 – A tumour composed of numerous thin-walled vessels located within the small intestinal submucosa (hematoxylin and eosin, 25×).

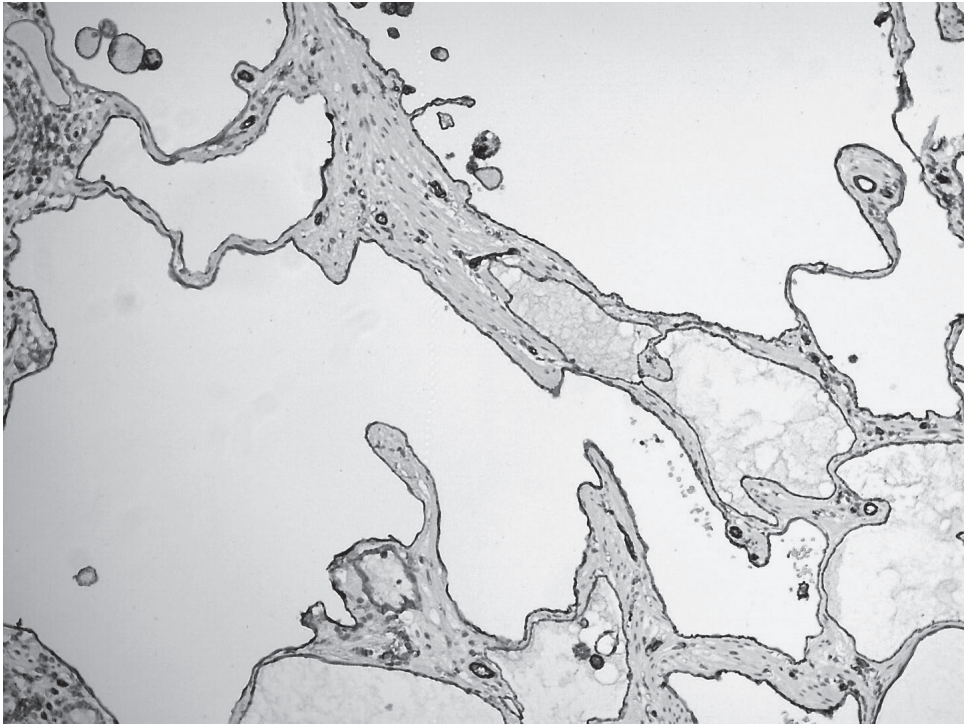


Figure 5 – Endothelial cells highlighted by the CD31 immunohistochemical stain (CD31, 100×).

for surgery. Laparoscopic exploration was performed, and surgeons found 8 cm long tumour between jejunum and ileum. Tumour was resected with LL enteroenteral anastomosis.

Pathohistological diagnosis was hemangioma (Figures 4 and 5).

This patient also recovered quickly after the surgery with normalisation of his hemoglobin and iron levels, with no more symptoms of fatigue and weakness.

Discussion

We present two rare cases of solitary small bowel hemangioma causing iron-deficiency anemia in two adult male patients. Small bowel hemangiomas are rare and account for 7–10% of all benign neoplasms of the small intestine (Takase et al., 2017; Otani et al., 2018). Most patients present with abdominal pain and intestinal bleeding with iron-deficiency anemia. Rarely patients can present with intussusception, obstruction or perforation. Tumours can be solitary and multiple as a part of the blue rubber-bleb syndrome, Maffucci syndrome and Klippel-Trenaunay syndrome. Its size may range from a few millimetres to several centimetres. Video capsule endoscopy has revolutionized the diagnostic approach to obscure gastrointestinal

bleeding (Quentin et al., 2007; Pinho et al., 2008). It is a noninvasive imaging test which can be recommended when the source of bleeding remains unidentified after upper and lower endoscopy. In our case VCE (video capsule endoscopy) found suspicious lesion of the small intestine in two patients with long standing anemia. Enteroscopy which is invasive procedure can be both diagnostic and therapeutic tool. In our case it demonstrated suspicious finding in the small bowel (Durer et al., 2018). Surgical laparoscopy with intestinal resection established the final histologic diagnosis and in the same time allowing definitive, curative treatment. Endoscopic interventions such as endoscopic mucosal resection (EMR), endoscopic sclerotherapy and argon-plasma coagulation (APC) are possible therapeutic options, but they might result in uncontrolled bleeding or perforation since intestinal hemangiomas originate from the submucosal layer and some of them are transmural. So endoscopic interventions are preferred when multiple and/or small hemangiomas are present.

We present these two cases to motivate colleagues gastroenterologist/endoscopist and surgeons to think of and recognize this rare condition. It is also important to consider carefully indications for possible endoscopic treatment.

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