

Case report

Rare case of successfully operated idiopathic colonic varicosity

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ARTICLE INFO

Keywords:

Idiopathic varicosity
Gastrointestinal bleeding
Laparoscopy
Case report

ABSTRACT

Introduction: Ectopic gastrointestinal varicosities are defined as dilated portosystemic collateral veins that may localize anywhere in the gastrointestinal tract outside the gastroesophageal region. Ectopic colonic varices can be considered idiopathic when other etiology that related to portal hypertension or portal vein thrombosis have been excluded.

Case presentation: A forty-five-year-old female patient has been under treatment for histopathologically confirmed ulcerative colitis since the age of 17. In her forties, the patient developed worsening hematochezia leading to severe anemia. Routine colonoscopy was performed which confirmed extensive rectosigmoid varices. A thorough investigation did not confirm any underlying causes, such as portal hypertension or cirrhosis.

Discussion: The selective percutaneous transhepatic mesenteric angiography, which is recommended as a diagnostic and therapeutic option, was not performed because the interventional radiologists did not consider embolization feasible. Laparoscopic rectosigmoid resection with high ligation of the inferior mesenteric vein led to complete remission of hematochezia. The final histological examination confirmed ectopic rectum and sigmoid varices, and ulcerative colitis was ruled out.

Conclusions: Lower gastrointestinal bleeding from the colonic varices is very rare, with only a few cases reported in the literature. In the idiopathic form, the prognosis is very good, given the absence of other underlying diseases causing portal hypertension. Ectopic varices present a clinical challenge as they are difficult to diagnose and localize. There are currently no clear guidelines for diagnosis and therapy, and recommendations are based on different case reports. Idiopathic cases can be treated effectively by resection of the affected bowel segment.

1. Introduction

This case report has been reported in line with the SCARE 2023 criteria. (1)

1.1. Prevalence and etiology of ectopic varices

Ectopic varices are defined as dilated portosystemic collateral veins located anywhere in the gastrointestinal tract outside of the esophageal and gastric regions (2). They most commonly develop in the duodenum, followed by the jejunum and ileum, and colon and rectum in terms of incidence (3). Sigmoid varicoceles are rarely mentioned in the literature and usually occur together with rectal varices (4). Colonic varicose veins account for only 0.07 % of all varicose veins in the gastrointestinal system, according to some authors. The incidence of the even rarer

idiopathic form is unclear (5,6).

Portal hypertension is the most common cause of portosystemic varices. Portal hypertension is usually caused by cirrhosis of the liver, which can have multiple causes, including viral or toxic liver damage, metabolic processes, or hepatocellular carcinoma. In addition to hepatic cirrhosis and hepatocellular carcinoma, secondary portal hypertension can also result from portal vein thrombosis. Varices in the gastrointestinal system can be triggered by hypercoagulopathy, myeloproliferative diseases, antiphospholipid syndrome, protein C, S or antithrombin III deficiency, or the Leyden mutation of factor V (7,8). Increased portal pressure, caused by liver cirrhosis or portal vein thrombosis, is the most common cause of GI varicosity. Less common causes in the colon may include congestive heart failure, local or systemic sepsis, mesenteric venous thrombosis, splenic vein thrombosis due to pancreatitis or tumors, and adhesions after surgery (9,10). These varicose veins in the

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<https://doi.org/10.1016/j.ijscr.2024.110196>

Received 11 June 2024; Received in revised form 15 August 2024; Accepted 17 August 2024

Available online 22 August 2024

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intestinal tract are called secondary ectopic varices, and they develop as a result of these underlying conditions. In addition to secondary forms, ectopic varicosity can also have an idiopathic form, in which the development of the disease cannot be explained by the aforementioned factors. Idiopathic colon varices are much rarer than secondary forms. Therefore, to establish a diagnosis of the disease, it is necessary to exclude the previously listed etiology factors behind the secondary form through evaluation (11–13).

1.2. Bleeding caused by ectopic varicosity

Ectopic varices account for approximately 2–5 % of gastrointestinal bleeding and may present as hematemesis or hematochezia, depending on their location in the intestinal tract. In the chronic form, they can cause iron deficiency anemia due to recurrent bleeding, while in the acute form, bleeding resulting from variceal rupture can lead to life-threatening shock. Norton et al. conducted a study on 169 patients who experienced variceal bleeding caused by ectopic varices. Bleeding in rare locations such as the vagina or ovary was reported in only a few percent of patients. Bleeding occurred in various locations among the patients, with the duodenum and jejunum/ileum being the most common sites (17 % each), followed by the colon (14 %), peristomal area (26 %), peritoneum (9 %), and rectum (8 %). Lebrek and Benhamou reported that 22 % of patients with ectopic varices experienced bleeding from the colon or rectum (14–16). Studies have shown that the risk of bleeding from ectopic varices is four times higher than from esophageal varices, and the mortality rate can be as high as 40 %. This is due to the sudden onset of bleeding with profuse melena or hematochezia in most patients with ectopic variceal bleeding. (3,17) Hematemesis is the most common symptom of duodenal varices.

1.3. Diagnosis and therapy of bleeding caused by ectopic varices of the lower gastrointestinal tract

The most common cause of lower gastrointestinal bleeding is colonic diverticulosis. Less commonly, it can be caused by various forms of IBD (ulcerative colitis, Crohn's disease), polyp or colon tumor, angiodysplasia, angioectasia, or hemorrhoidal nodules. Less than 1 % of colonic bleeding can be attributed to the occurrence of idiopathic varices. In the case of secondary forms, hemorrhoids and rectal or sigmoid varices co-occur in about 30 % of cases, as confirmed by Hosking et al. in their study involving 100 patients suffering from liver cirrhosis. However, there is no clear correlation for idiopathic forms. Lower gastrointestinal bleeding is initially evaluated by upper and lower endoscopy. Most of the time, it is the first line of investigation. In patients who do not have portal hypertension and the source of the gastrointestinal bleeding cannot be clearly identified by gastroscopy or colonoscopy, the presence of ectopic varices should always be considered (18–22). Endoscopy provides both diagnostic information and therapeutic options. There are several endoscopic options available to stop the bleeding or to prevent further bleeding, such as injection sclerotherapy, band ligation, clipping, and argon plasma coagulation. The use of tissue adhesives could be helpful when adequate visualization is not possible. Endoscopy is also beneficial to follow up on therapy of the varix after intervention (24,25).

In addition to colonoscopy, selective mesenteric angiography is an effective method for diagnosing colonic varices, which also offers an opportunity for therapeutic intervention (10). Angiography can be selectively performed transhepatically through the portal vein (if there is no portal thrombosis) or transjugularly by catheterizing the mesenteric veins. This procedure enables the localization of colonic varices and the identification of the source of gastrointestinal bleeding; moreover, selective embolization can be performed in one session to stop the bleeding. Variceal embolization is accomplished by occluding the vein supplying blood to the bleeding area. Embolization of the veins draining into the ectopic varices can be done with coils, 100 % alcohol, or Gelfoam (Pfizer, New York, NY) or a combination of these (8,25).

Mesenteric angiography is an excellent diagnostic tool, but some authors have suggested that diagnostic errors can occur with this method. Varices may be detected only during the venous phase of angiography, and the amount of contrast material may also be insufficient to detect varices (27,28). Also selective embolization in cases of secondary forms, the likelihood of recurrence and re-bleeding within one year is very high due to persistent portal hypertension (29).

In the treatment of secondary forms, portal decompression can also yield results if endoscopic hemostasis or embolization has failed. Transjugular intrahepatic portosystemic shunt (TIPS) can be successfully used in the treatment of secondary ectopic variceal bleeding. However, depending on the degree of portal hypertension and the cause of its development, this procedure carries the risk of hepatic decompensation and encephalopathy. The procedure is contraindicated in the case of a high Child-Pugh score, advanced heart or renal failure, and cannot be performed in the presence of portal vein thrombosis. After the intervention, we also have to consider the occlusion of the stent, and reintervention may also be necessary in the case of splenorenal or gastrotorenal shunt (30–32).

In cases where endoscopic and interventional radiological procedures are not successful in controlling the bleeding, then a surgical solution should be chosen, which usually means resection of the affected intestinal segment (33–35).

1.4. Idiopathic ectopic varicosity

The management of ectopic varices is a difficult and often multidisciplinary challenge. It requires a team consisting of a hepatologist, endoscopic gastroenterologist, interventional radiologist, and surgeon, all to establish the correct diagnosis and select the appropriate therapy. Idiopathic colonic varices can be diagnosed after exclusion of underlying liver disease and portal hypertension. Recognizing this condition is also very important because, in the absence of liver disease, the prognosis is much better than in the case of secondary forms, and it can be cured by resection of the affected intestinal segment (11–13,33,34). Ectopic varices in the lower gastrointestinal tract are most commonly formed in the cecum and rectum (36).

2. Case presentation

A forty-five-year-old female patient has been under treatment for histopathologically confirmed ulcerative colitis since the age of 17. In December 2022, she was admitted to the Intensive Care Unit due to hematochezia which had been present 4–5 times a day for months and consequent severe microcytic anemia. On arrival at the hospital, the hemoglobin value was 59 g/L, the red blood cell count was 4.00 T/L and the hematocrit value was 112 g/L. After administration of 4 units of blood transfusion, she was admitted to the Gastroenterology department with blood count values in the normal range. After that, during the comprehensive examination, until surgery, the control blood count values remained within the normal range that did not require transfusion. The patient did not mention any pain, fever, recent weight loss, gynecological or urological problems. Regarding her personal habits, she denied regular alcohol consumption and smoking, as well as drug consumption. Family history was negative for colorectal cancer as well as for previous occurrence of gastrointestinal bleeding. She denied abdominal pain, weight loss, fatigue, or fever. On physical examination, besides an anemic appearance, there were no abdominal tenderness and no palpable signs of tumor. Her laboratory findings yielded severe hypochromic microcytic anemia, low ferritin levels with normal liver enzymes, folic acid, B12 levels, and a markedly prolonged INR and APTT values. Imaging studies (abdominal ultrasound, abdominal CT) ruled out the presence of any tumor or diverticulosis. Gastroscopy confirmed bile reflux, gastritis, and duodenitis which did not explain the hematochezia and severe anemia. Considering the persistent and worsening hematochezia despite therapy for ulcerative colitis, colonoscopy was

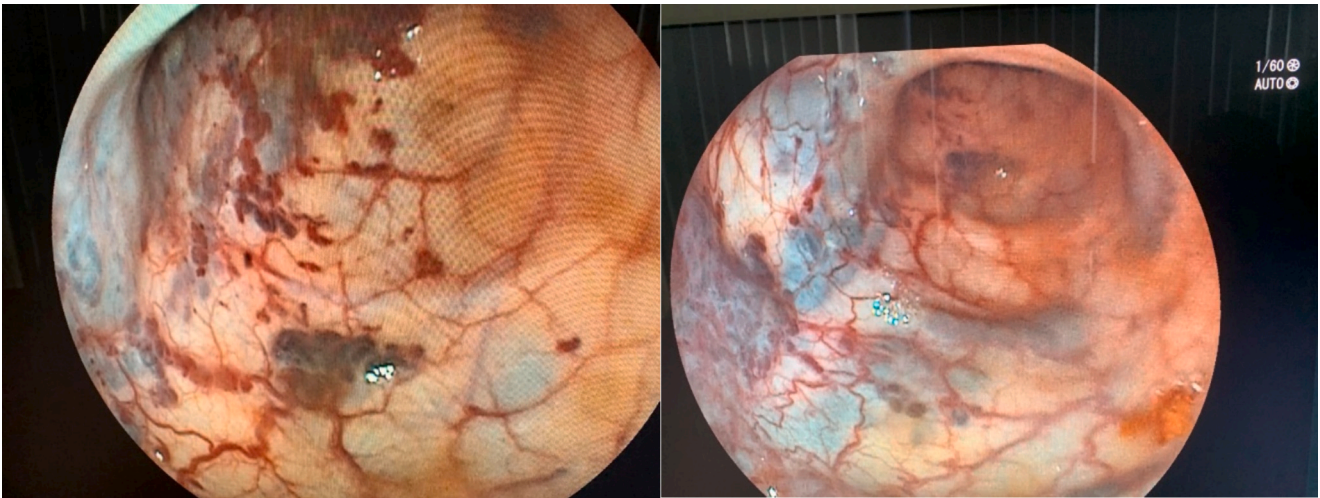


Fig. 1. Colonoscopy picture: tortuous dilatated veins in the sigmoid colon with disappearance of haustration.

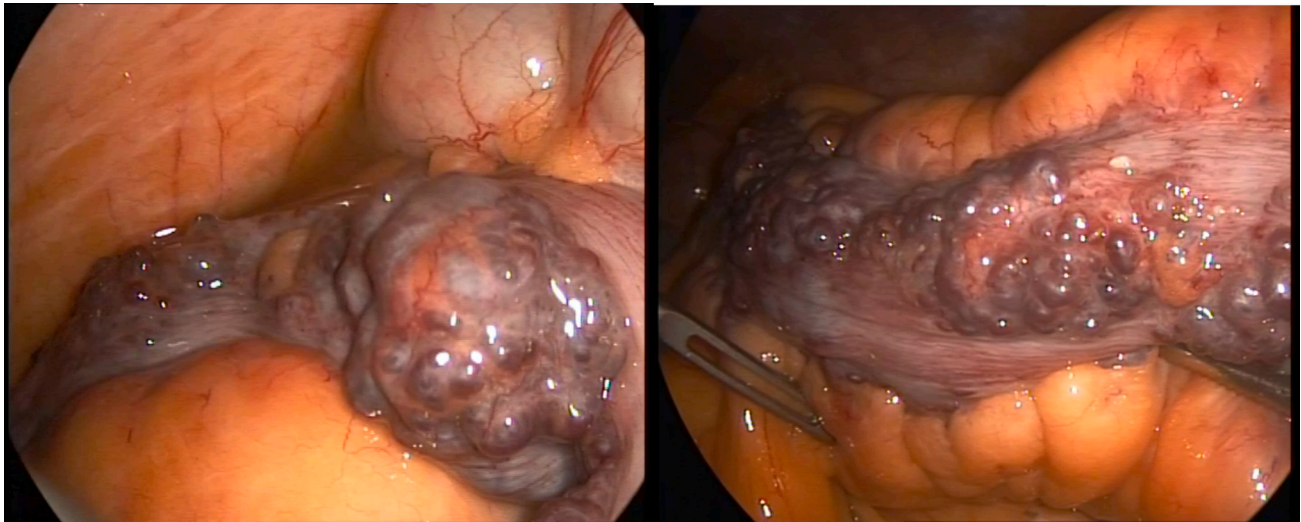


Fig. 2. Intraoperative picture of the deformed colon by large varicose veins.

performed.

During the colonoscopy, dilated submucosal veins were found from the sigmoid bowel to the anus, and the disappearance of the haustration. Given the colonoscopy findings, biopsy was not performed due to the risk of bleeding from the varices.

CT angiography and MR examination described the portal vein as 15 mm in diameter at the portal region, but distally up to the superior mesenteric vein at the level of the pancreatic corpus it was found to be significantly dilated (45 mm in diameter). The entire length of the portal vein was homogeneously filled, and there were no signs of thrombus. No abnormalities were seen in the mesenteric veins except for some small calcification. Abdominal Doppler ultrasound revealed normal portal flow volume, and acoustic radiation force impulse (ARFI) elastography did not show liver fibrosis. As the patient had no radiomorphological evidence of liver cirrhosis, splenomegaly, pancreatic lesions, or portal vein thrombosis, nor any evidence of hepatocellular carcinoma, the colonic varices were considered idiopathic. Liver enzyme values were repeatedly checked, and tumor marker levels were within the normal range. Additionally, serological markers for hepatitis B and C ruled out viral hepatitis.

Subsequently, due to recurrent hematochezia and anemia, another colonoscopy was performed which confirmed the previous diagnosis and

described tortuous venous dilation from the sigmoid colon to the anal ring, with disappearance of haustration (Fig. 1). Superficial biopsy was also performed but the histopathology examination ruled out the diagnosis of IBD.

Selective mesenteric angiography is used for both diagnostic and therapeutic purposes. Variceal embolization is accomplished by occluding the vein supplying blood to the bleeding area, which may be a useful interventional approach to avoid surgery. Unfortunately, interventional radiological consultation considered embolization not feasible due to known vena portae dilatation and the large diameter of the patient's ectopic varices. With no further diagnostic or treatment options available, we decided on a surgical exploration.

Accordingly, in the beginning of 2023 a laparoscopic rectosigmoid resection was performed at our department of surgery. During the surgery we found that from the sigmoid-descending border the colon was deformed by large varicose veins all the way to the upper third of the rectum (Fig. 2). Proximally the colon was completely intact (Fig. 3). The mesosigmoid and mesorectum was also full of thick, dilated, tortuous veins (Fig. 4). The surgical and endoscopic findings were correspondingly consistent, and therefore, laparoscopic rectosigmoid resection with end-to-end instrumental descenderectostomy with high ligation of the inferior mesenteric vein was performed. The removed specimen

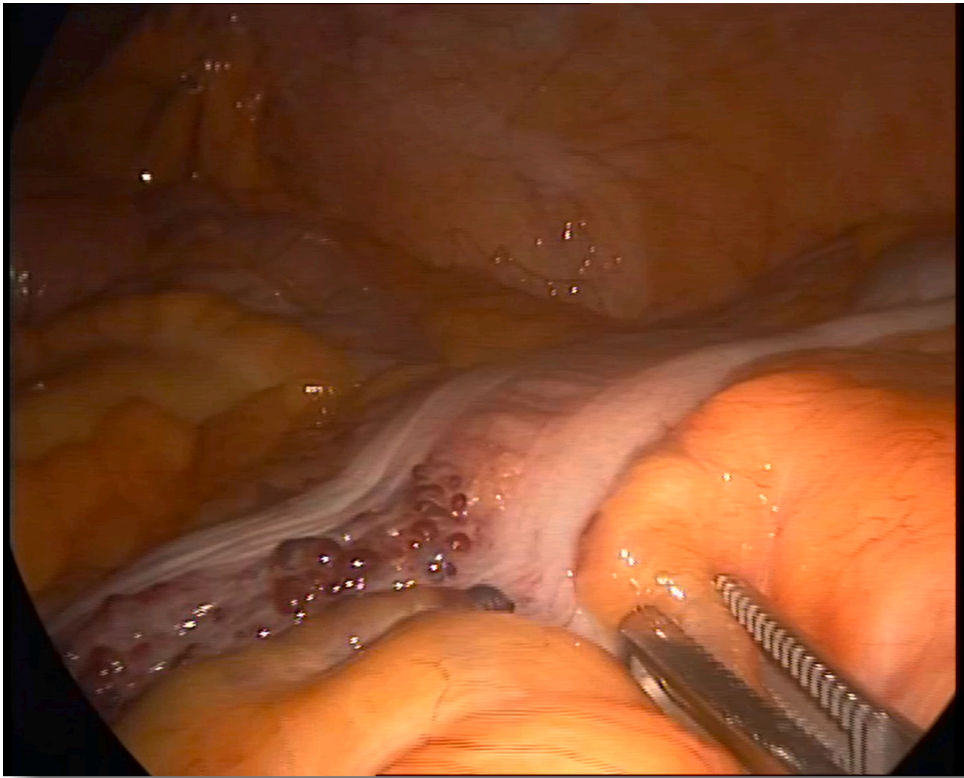


Fig. 3. Intraoperative picture of the colon: proximally from the sigmoid-descending border the colon is completely intact.

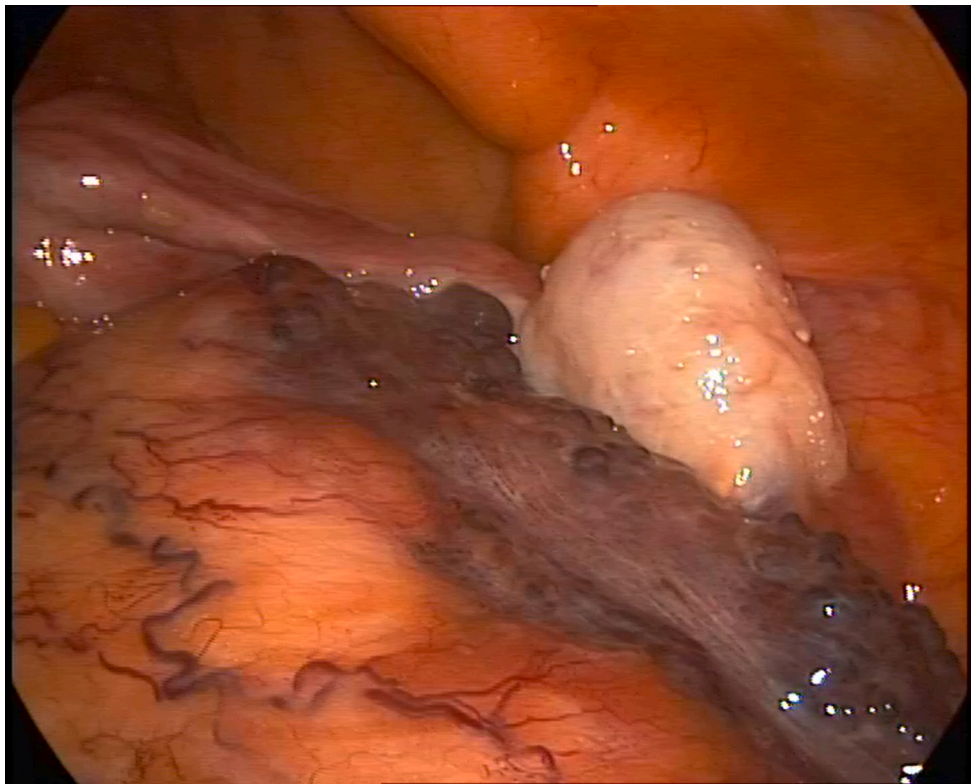


Fig. 4. Intraoperative picture of the mesosigmoid and mesorectum with thick, dilated, tortuous veins.

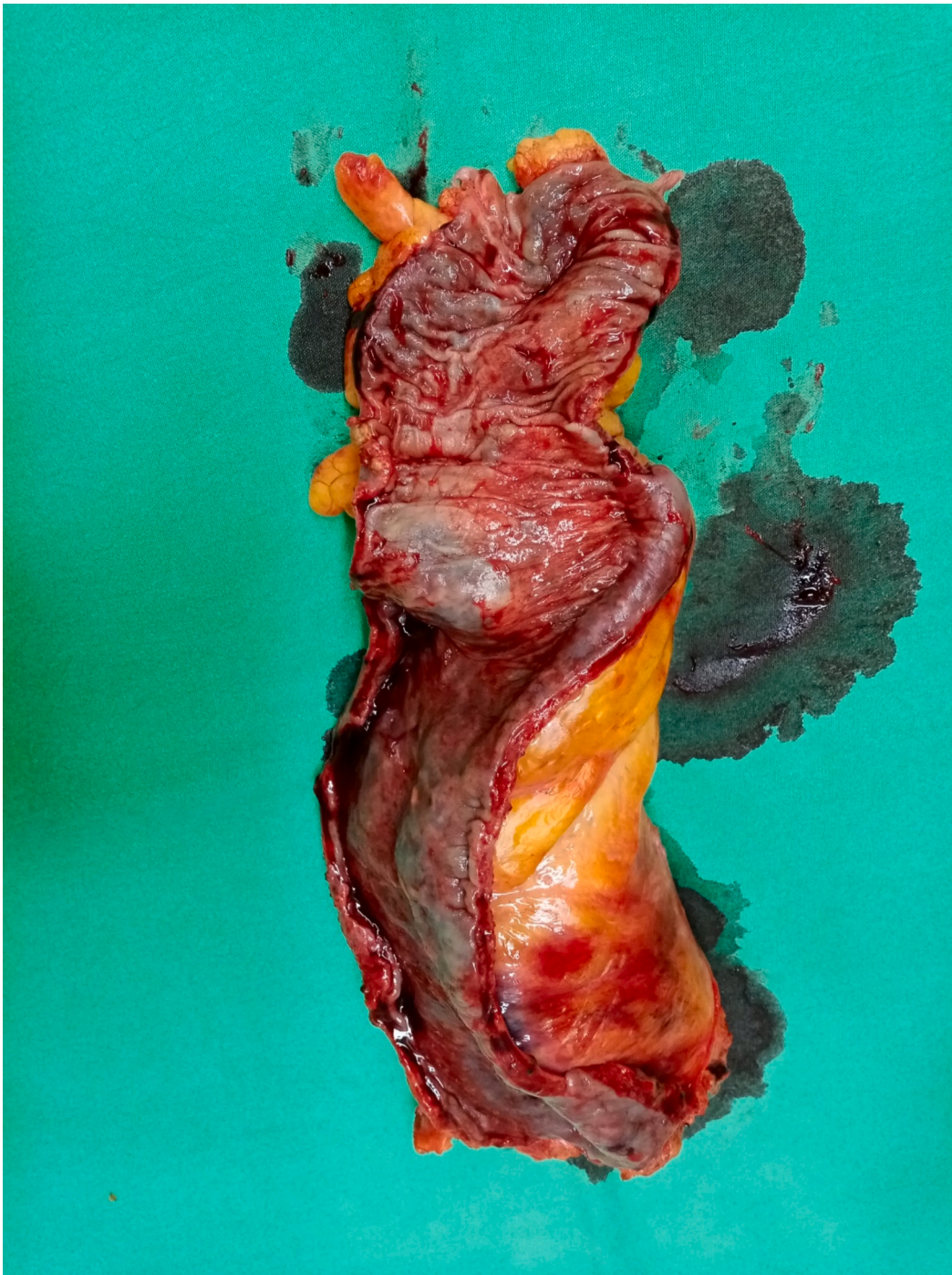


Fig. 5. The resected rectosigma.

(Fig. 5). In the postoperative period, we used ulcer and thrombosis prophylaxis, mobilized her, and gradually increased her nutrition parenterally and then orally. Resumption of bowel function was on the third day after surgery, when the drain was removed and oral feeding with a pasty diet was started. The patient was discharged home on the postoperative seventh day, without complications. The final histological examination confirmed ectopic rectum and sigmoid varices, and ulcerative colitis was again ruled out.

During the follow-up period, the patient no longer experienced hematochezia or transrectal bleeding. Her blood count was normalized in her control labs. A control colonoscopy performed after surgery did

not described tortuous venous dilation on the remaining colon section (Fig. 6).

3. Discussion

The presented patient has been treated for ulcerative colitis since childhood. In her forties, the patient developed worsening hematochezia leading to severe anemia. A colonoscopy at the age of 17 showed no colonic varices. Due to increasingly frequent hematochezia, another examination was performed years later which confirmed extensive rectosigmoid varices. The appearance of colonic varices in endoscopic

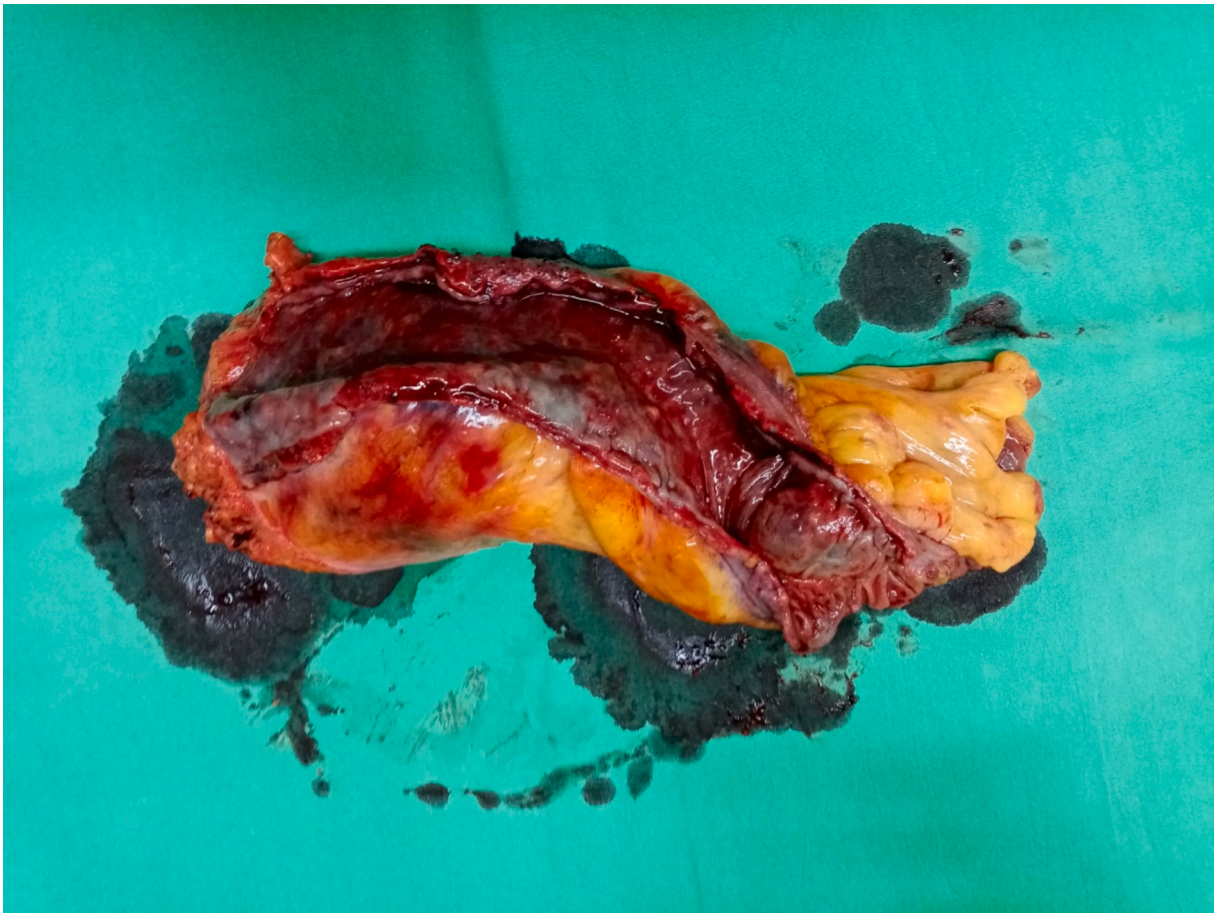


Fig. 5. (continued).

examination was quite typical and the diagnosis was easily made.

CT angiography followed by an MR scan showed the vena portae to be significantly dilated at the level of the pancreas corpus. Since CT angiography, MR, abdominal Doppler, and acoustic radiation force impulse (ARFI) elastography did not show liver fibrosis, portal vein thrombosis, or hepatocellular carcinoma, the superior mesenteric vein dilatation was considered to be congenital. Accordingly, ectopic colonic varices diagnosed during a later colonoscopy can also be considered idiopathic. The other less frequent causes responsible for the development of portal hypertension (heart failure, pancreatic diseases, malignant tumors, sepsis, hematological diseases) were not supported by clinical, physiological, or imaging data.

Colonic varices are most commonly diagnosed by routine or emergency colonoscopy in search for the cause of a lower gastrointestinal bleeding. Nevertheless, it is important to differentiate between rectal varices and hemorrhoids, as their therapeutic consequences differ. Rectal varicosity extends above the level of the levator ani muscle and is dilated in the form of tortuous 3–6 mm diameter submucosal dark blue veins that do not prolapse in front of the rectoscope during examination (18,19). Colonoscopy will often show the colonic varices, but in some cases, due to hypotension caused by gastrointestinal bleeding, varices collapse and cannot be visualized properly during colonoscopy. Also, we should be aware that even in normotensive patients, the intraluminal pressure caused by insufflation compresses and flattens the dilated veins. Since lower gastrointestinal bleeding caused by ectopic varices is rare, the examiner does not expect it, so it can easily be mistaken for polyps, some forms of inflammatory bowel disease, or cancer. In such cases, severe bleeding may occur during biopsy sampling. (24,25) Another effective diagnostic modality is selective mesenteric angiography, which can also be a therapeutic intervention, as well (5,9,11).

The patient presented here has undergone three colonoscopies in her lifetime. During the first examination they found no dilated veins in the rectosigmoid segment, the diagnosis was made only much later. The diagnosis of idiopathic ectopic varices may require multiple imaging studies and repeated colonoscopies because the development of colonic varices in the absence of portal hypertension or portal vein thrombosis is considered a rare finding. In rare idiopathic forms, embolization may provide a definitive solution. The selective percutaneous transhepatic mesenteric angiography, which is recommended as a diagnostic and therapeutic option, and can help avoiding surgery, was not performed because the interventional radiologists did not consider embolization feasible due to known vena portae dilatation. Another reason why it was not performed is that sometimes the result of this procedure is unsatisfactory due to the large diameter of the patient's ectopic varices, which leads to the failure of embolization and can cause repeated bleeding. (11,29) Endoscopy also provides therapeutic features such as injection sclerotherapy, band ligation, clipping etc. Due to the amount and size of the dilated veins, we considered it more effective in terms of therapy to remove the affected colon section.

Idiopathic cases, in the absence of portal hypertension and liver cirrhosis, have a much better prognosis than secondary cases and can be treated effectively by resection of the affected bowel segment (11–13,33,34). In our case, laparoscopic rectosigmoid resection with high ligation of the inferior mesenteric vein led to complete recovery. There was no further bleeding, and the patient's blood count resolved spontaneously. Colonoscopy performed 6 months after surgery no longer showed varicosity. It is important to note that the biopsy taken during the last colonoscopy and the definitive histology of the resected rectosigmoid section did not confirm ulcerative colitis, so it is possible that the patient's initial complaints were also caused by symptoms

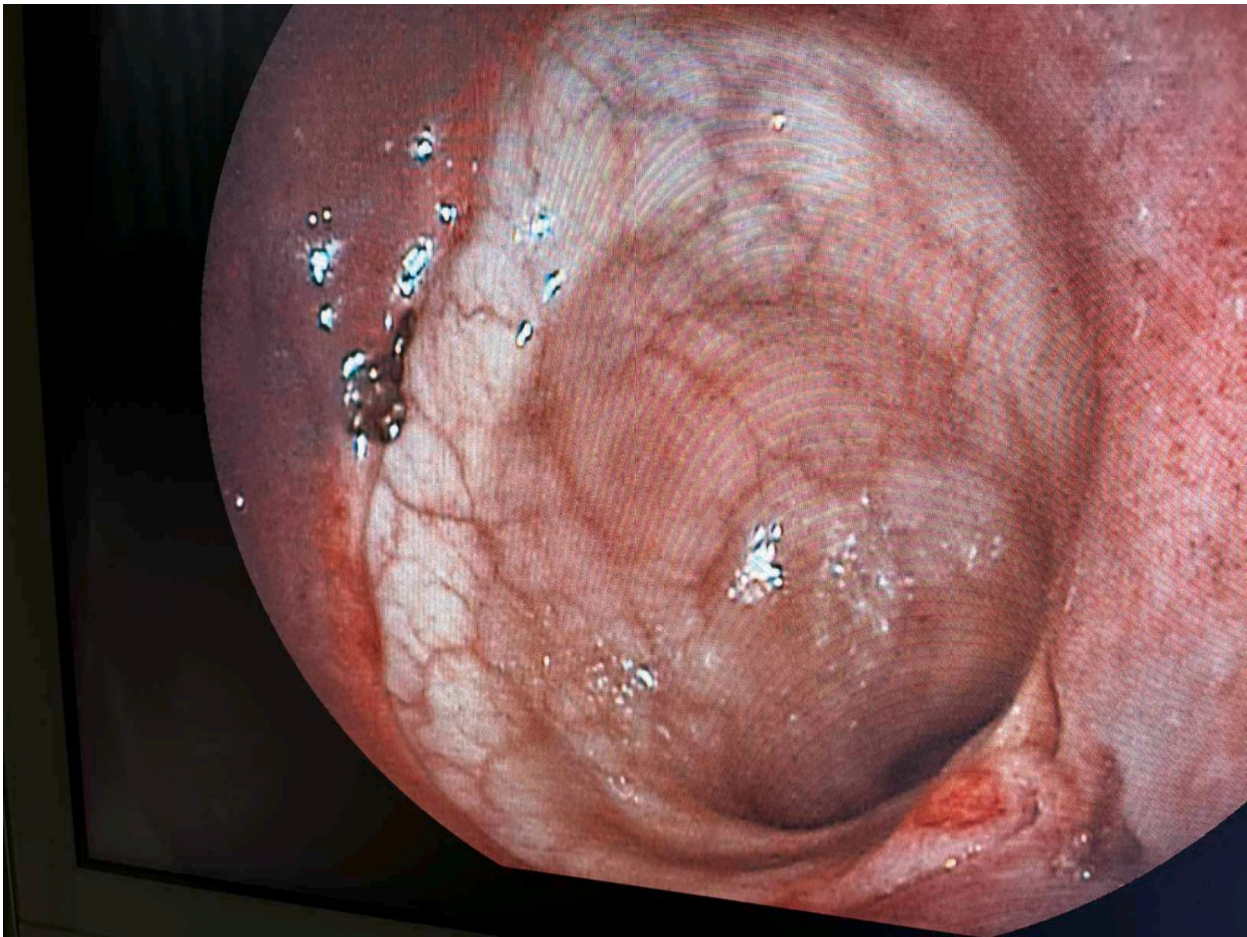


Fig. 6. The colonoscopic finding after surgery.

arising from the venous dilation.

4. Conclusions

The clinical relevance of this case is that in the absence of portal hypertension and liver cirrhosis, it is a challenging diagnostic decision to conclude that idiopathic colon varicosity may be behind the complaints of a patient, and therefore diagnosis and treatment may be delayed for years. Further case-studies are needed to develop standard treatment strategies by comparing the efficacy and safety of treatment options. The precise early diagnosis is important because selective embolization or laparoscopic resection can achieve a complete cure, preventing the complications mentioned above or a possible mistreatment (11–13,33,34).

Ethical approval

The case report was approved by *Regional Institutional Research Ethics Committee, Clinical Center, Undercity of Debrecen* (6831–2024).

The patient consented to the publication of the study, as the study does not contain data on the individual identification or ethnic identification of the patient.

Funding

The study sponsor has no involvement in data collection and analysis, and in the writing of the manuscript.

Author contribution

Gergely Kóder contribute to the publication.
 Tamás Dinya contribute to the publication.
 László Damjanovich contribute to the publication.
 Dezső Tóth contribute to the publication.
 Lóránt Ágoston contribute to the publication.
 Miklós Tanyi contribute to the publication.

Guarantor

The Guarantor is the corresponding author, Gergely Kóder.

Research registration number

Not applicable.

Declarations

The work has been reported in line with the SCARE criteria.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Content for publication

The manuscript does not contain any individual personal data in any form.

Declaration of competing interest

The study sponsor (University of Debrecen, Hungary) has no involvement in data collection and analysis, and in the writing of the manuscript.

Data availability

All the datasets used in this report about the patient can be found in the digital register of the Surgical Department of the University of Debrecen.

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

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