CLINICAL CASE

MESO-ILEAL ECTOPIC VARICOSE VEIN WITH UTERINE SYSTEMIC SHUNT AS AN UNCOMMON CAUSE OF SEVERE LOWER GASTROINTESTINAL BLEEDING IN PATIENT WITH MILD PORTAL HYPERTENSION

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Abstract

Ectopic varicose veins of the small intestine are an uncommon cause of gastrointestinal bleeding in patients with portal hypertension. We present the case of a 52-year-old female patient with mild portal hypertension and repeat episodes of lower gastrointestinal bleeding because of meso-ileal ectopic varicose vein with systemic uterine shunt secondary to previous pelvic surgery, radiological findings and treatment performed.

Keywords: gastrointestinal hemorrhage, varicose vein, systemic uterine shunt, portal hypertension, computed tomography angiography.

Introduction

Lower gastrointestinal bleeding secondary to ectopic varices in patients with mild portal hypertension without other stigmata of chronic liver disease is a rare scenario. The development of a meso-ileal ectopic varicose vein with uterine systemic shunt secondary to previous pelvic surgery for removal of an intrauterine device in a patient with mild portal hypertension without oesophagogastric varices and which causes bleeding in the transfusion range is a form of bleeding not described in the literature, so we present this case for its originality and relevance in the form of presentation, management and treatment.

For this reason, we present the clinical case of a 52-year-old patient with the previously described background to visualise this unpublished case, providing the clinical management and diagnostic imaging where the varicose vein was visualised and the endovascular treatment that allowed its embolisation in a safe and effective manner.

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Clinical case

52-year-old woman, smoker, drinker and with a previous history of three pregnancies (two vaginal deliveries and one caesarean section). She underwent surgery due to a complication in the removal of an intrauterine device (Essure®) with salpinguectomy and partial hysterectomy four years ago.

She came to the emergency department for intermittent rectorrhagia of three days duration, although on the last day it was continuous with clots. She presented with mucocutaneous jaundice, stigmata of chronic liver disease with telangiectasias and palmar erythema, hepatomegaly without signs of ascites, hypotension of 77/66 mmHg well tolerated with a heart rate of 100 bpm and a positive rectal examination for red blood.

Complete blood count of 1.24x 10^6 µ/L (reference 4.20-6.10 µ/L), haemoglobin of 4.5 g/dL (reference 12-16 g/dL) with normal urea and activated partial thromboplastin time of 24.9 s (reference 26-39 s).

Biochemistry showed total bilirubin of 2.40 mg/dL (reference 0.3-1.20 mg/dL), GGT of 404 U/L (reference 5-38 U/L), ALT of 53 U/L (reference 10-49 U/L) and alkaline phosphatase of 667 U/L (reference 46-116 U/L).

Upper gastrointestinal endoscopy was performed and no oesophagogastric varices or other potentially bleeding lesions were identified. Colonoscopy was also performed with no relevant findings. Abdominal ultrasound showed signs of chronic liver disease and mild ascites. This episode resolved with transfusion of red blood cell concentrates.

Twelve days later, she came to the emergency department for a new episode of rectorrhagia with severe anaemia in transfusion range. Contrast-enhanced magnetic resonance enterography showed a hypervascular structure with submucosal arrangement in the wall of the pelvic ileum communicating with the prominent superior mesenteric vein and with right periuterine branches **(Figure 1)**, and subsequently non-contrast and contrast-enhanced computed tomography of the abdomen and pelvis in arterial and venous phases, which corroborated the vascular finding **(Figure 2)**.

To confirm portal hypertension and the findings of the imaging tests, manometry of the suprahepatic veins was performed through the right internal jugular vein, obtaining a transhepatic gradient of 8 mmHg compatible with mild portal hypertension. Venography of the ovarian and hypogastric veins was also performed, without identifying any communication between them and the ileal veins. Subsequently, indirect portography was performed by catheterising the superior mesenteric artery, showing a dilated ileal vein in the venous phase that reached the wall of an ileal loop and opacified part of the uterine body and uterine veins.

In a second stage, direct portography was performed by transhepatic puncture with a 21G needle through a 6F introducer, 4F catheter and Terumo® hydrophilic guide, and the distal ileal branch was catheterised with venographic series confirming the presence of dilated ileal varicose veins with fistula to the uterine veins and early opacification of both hypogastric veins. These branches were selectively catheterised with a Marathon® microcatheter and embolised with Onyx® 34. In the final control, embolisation of the branches at the level of the loop and the origin of the fistula with the uterine veins is observed **(Figure 3)**. Finally, the transhepatic tract was embolised with Espongostan® Film. More than six months after the procedure, the patient has had no new episodes of lower gastrointestinal bleeding.

Discussion

Ectopic variceal bleeding in patients with portal hypertension is rare and few cases have been reported in the literature. In the published cases, the most common is the history of an abdominal surgical procedure that has triggered the formation of new spontaneous and ectopic portosystemic shunts through postoperative scar tissue, with enterectomy being the most commonly described procedure[1]. The development of a meso-ileal ectopic variceal shunt with systemic-uterine shunt secondary to a previous surgical procedure of salpinguectomy and partial hysterectomy leading to repeated lower gastrointestinal bleeding and its diagnosis and treatment has not been described.

Gastrointestinal bleeding is defined as blood loss from the digestive tract and may constitute an emergency requiring urgent admission in order to treat or prevent hypovolemic shock, identify the bleeding lesion and treat it for haemostatic or curative purposes[2,3].

Oesophagogastric varices are the most frequent cause of upper gastrointestinal bleeding in patients with liver cirrhosis and are a consequence of the development of portal hypertension, with a risk of bleeding when the pressure gradient exceeds 12 mmHg.

Cirrhotic patients with portal hypertension may also develop varices in other locations. These are called ectopic varices and are defined as portosystemic venous collaterals that can occur anywhere except in the cardio-oesophageal

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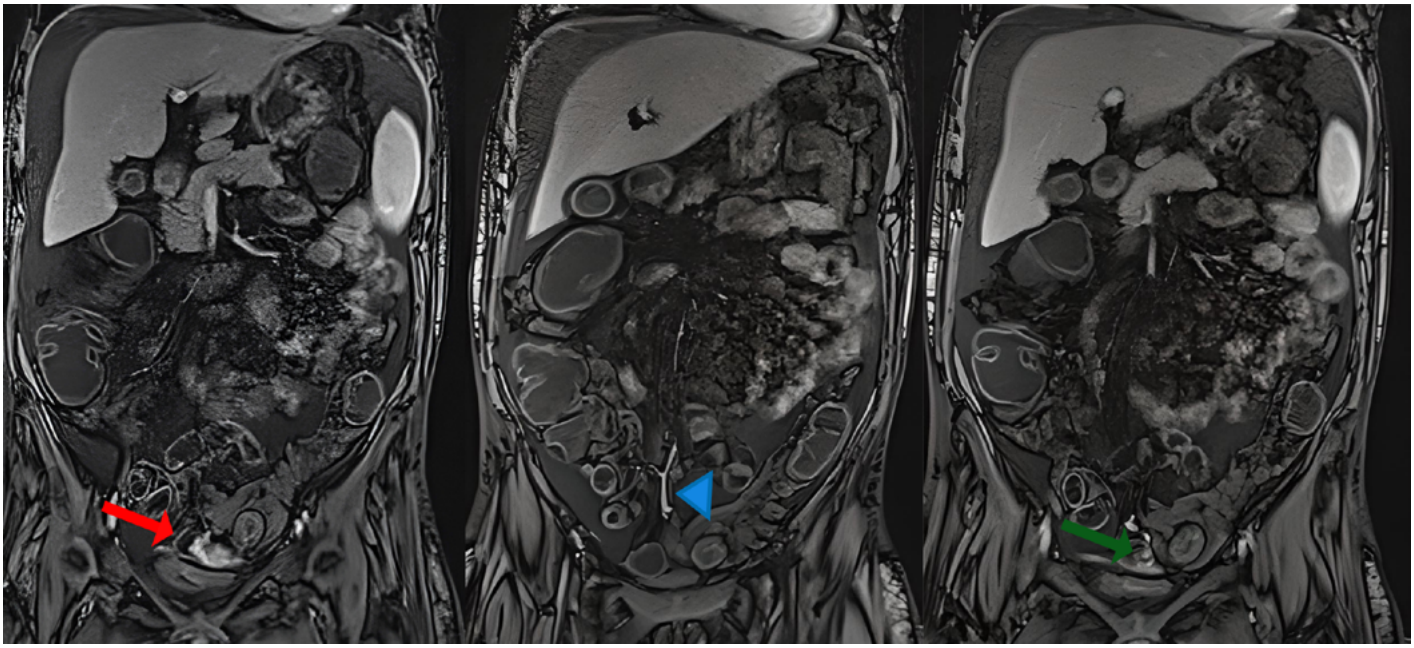

Figure 1. Magnetic resonance enterography T1 sequences with fat saturation and contrast, showing a hypervascular structure with submucosal disposition in the wall of the pelvic ileum (red arrow) that communicates with the prominent superior mesenteric vein (blue arrow) and with periuterine branches (green arrow).

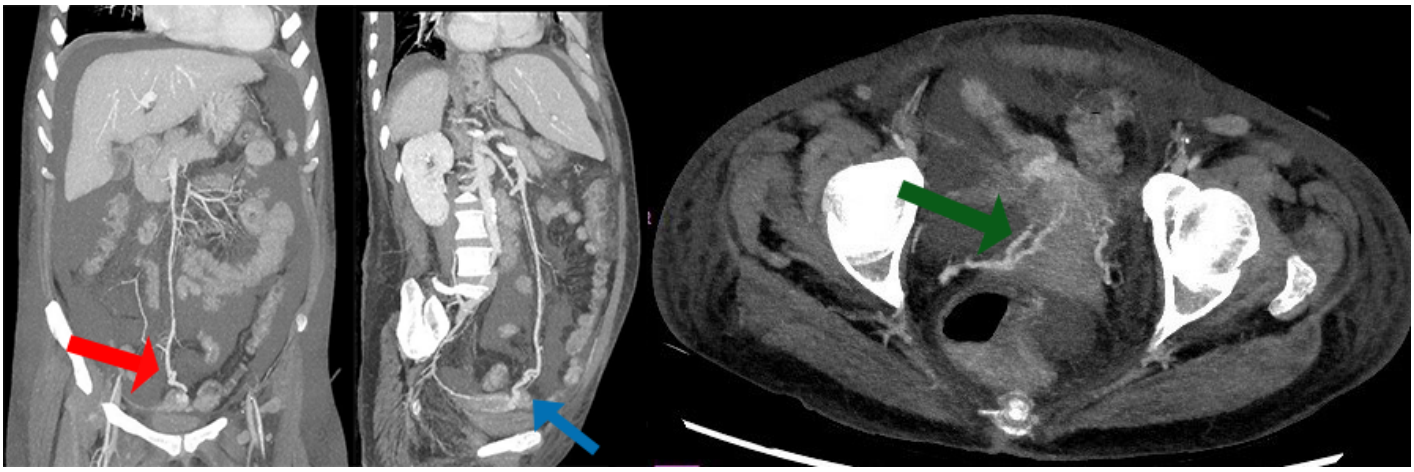

Figure 2. Computed tomography angiography of the abdomen and pelvis, maximum intensity projections with contrast and in the venous phase, in coronal, oblique sagittal, and axial planes. Ectopic meso-ileal varix with systemic-uterine shunt. Enlarged superior mesenteric vein continuing into the ileal vein (red arrow) protruding over the submucosa of the pelvic ileum (blue arrow) and opacifying the uterine veins (green arrow).

region[4]. Ectopic varices include isolated type II gastric varices (antrum, body and around the pylorus), those of the abdominal wall and peritoneal space. It has been reported that up to 8% of patients with portal hypertension may present them, with the most frequent location being the duodenum followed by the jejuno-ileum, colon, rectum, biliary tree, ovarian circulation and peritoneum[5].

They can develop anywhere in the intestinal or biliary circulation as a result of short circuits of the portal system due to high pressure in this system, because this hypertension and congestion in the splanchnic circulation increase the susceptibility to damage by impaired oxygenation and ischaemia, being greater in portal hypertension of extrahepatic origin[6].

In patients without portal hypertension, the occurrence of ectopic varices has been explained in several ways. Firstly, it has been suggested that they may be related to intestinal surgeries in which systemic drains are anastomosed to portal draining structures. In strictures of intestinal surgeries or in adhesion tissues, arteriovenous fistulas secondary to trauma are also thought to be factors facilitating the development of ectopic varices. The risk of developing ectopic varices is higher if there is a surgical history, whether or not it involves the portal circulation, stenosis of the stoma, or in patients with a

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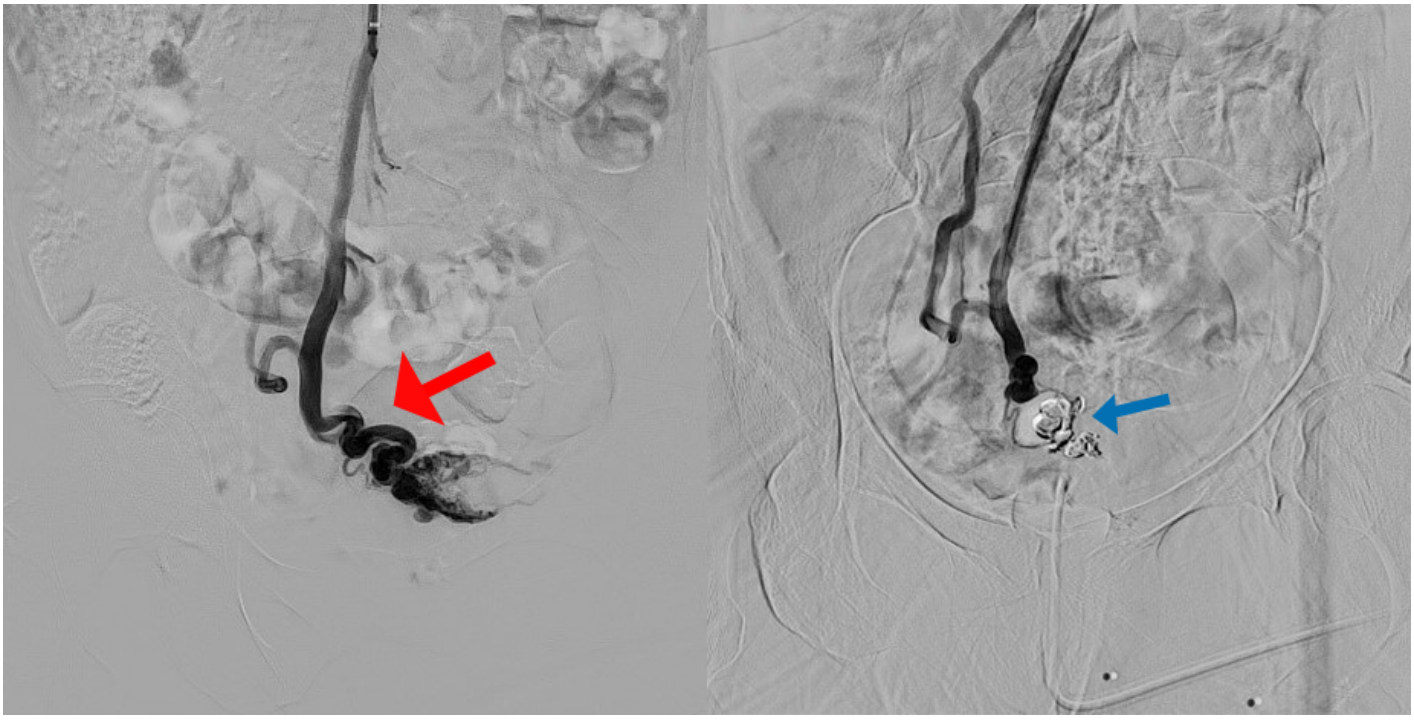

Figure 3. ***Digital subtraction angiography. Meso-ileal varix with systemic-uterine shunt (red arrow) and subsequent embolization with Onyx® (blue arrow) .***

surgical history who subsequently develop cirrhosis and portal hypertension. The likelihood of developing bleeding is greater the larger the size of the ectopic varices[7].

Contrast-enhanced computed tomography is a good alternative for the diagnosis of intestinal varices[8]. Radiological findings of small bowel varices have been rarely described in the literature. The presence of an intestinal intramural lesion with no enhancement in the arterial phase and homogeneous enhancement in the venous phase in a patient with intestinal bleeding should raise suspicion. Differential diagnosis should be established mainly with small bowel tumours, including haemangioma and GIST (gastrointestinal stromal tumours). Therapeutic possibilities include percutaneous embolisation and surgery[9].

Endovascular treatment of bleeding from ectopic varices allows surgery to be dispensed with, avoiding major bowel resections, and is a minimally invasive curative technique.

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