

Vascular malformation of the jejunum presenting as obscure gastrointestinal haemorrhage: detection with multi-detector CT angiography

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ABSTRACT

Vascular malformation of the small bowel is an uncommon cause of gastrointestinal haemorrhage. Phlebectasia or venous ectasia is a rare benign vascular anomaly of the gastrointestinal tract. We report a 39-year-old Egyptian man presenting with multiple jejunal phlebectasia, liver haemangioma and port-wine naevus. Despite recurrent melaena, the results of various routine investigations, including repeated endoscopic procedure, were negative. The site and aetiology of bleeding was detected using multidetector computed tomography angiography (MDCT) and was further confirmed by double-balloon enteroscopy. This report emphasises the potential of MDCT angiography in the diagnosis of small intestine lesions presenting as obscure bleeding.

Keywords: jejunum, MDCT angiography, obscure gastrointestinal haemorrhage, phlebectasia, vascular malformation

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INTRODUCTION

Vascular malformation of the small bowel is an uncommon cause of gastrointestinal haemorrhage.⁽¹⁾ Most vascular lesions of the small bowel present as chronic gastrointestinal bleeding. The diagnosis of small bowel lesions is challenging if the upper gastrointestinal endoscopy and colonoscopy results are negative.⁽²⁾ Multidetector computed tomography (MDCT) angiography is proving to be a very robust tool in identifying the source of gastrointestinal bleeding.⁽³⁾ We describe a case of jejunal phlebectasia presenting as recurrent melaena and anaemia, with emphasis on the role of MDCT angiography in the diagnosis.

CASE REPORT

A 39-year-old Egyptian male patient presented with a



Fig. 1 Photograph of the anterior abdominal wall shows a reddish-to-purplish flat lesion (arrow), suggestive of a port-wine stain.

history of recurrent painless melaena for the past 15 years. On numerous previous hospital admissions, he had received multiple units of blood transfusion for anaemia. The reports of previous upper endoscopy and colonoscopy examinations were normal. About eight years ago, the patient had presented with massive bleeding, and exploratory laparotomy was performed. Multiple prominent veins were noted over the surface of the jejunum. Intraoperative enteroscopy was not available. No definite bleeding site was identified and no surgical resection was performed. Clinical examination of the patient at the present admission was normal except for a port-wine naevus noted on the anterior abdominal wall (Fig. 1). Laboratory investigations showed a low haemoglobin concentration (Hb 9 g/dl; normal 12–16 g/dl) level. Biochemical parameters, including electrolytes, liver and renal function tests, were within normal limits. Repeat upper gastrointestinal endoscopy

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Fig. 2 Multidetector CT angiogram coronal maximum-intensity-projection image from the venous phase clearly shows multiple tortuous venous channels in the wall of the jejunal loops (arrowheads).

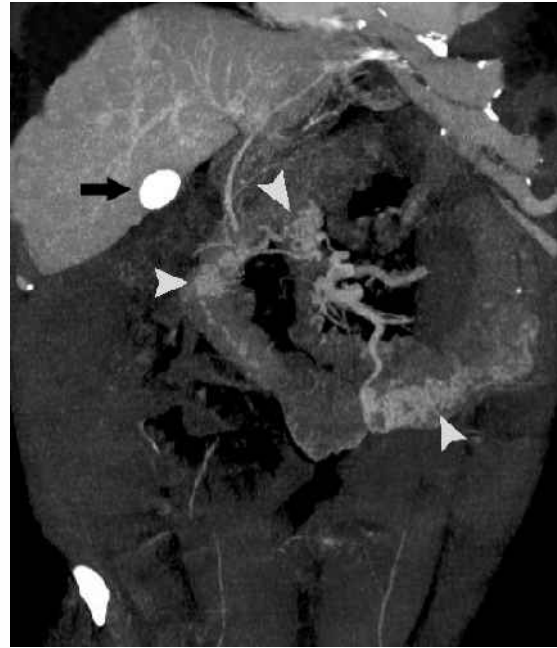


Fig. 3 Multidetector CT angiogram coronal maximum-intensity-projection image from the venous phase (section posterior in the previous image) shows a gall bladder calculus (black arrow) in addition to venous channels (arrowheads).

and colonoscopy examinations were unremarkable. Abdominal ultrasonography revealed multiple small liver haemangiomas and a gall bladder calculus.

MDCT angiography was performed on a 64-channel multidetector scanner. For small bowel distension, 900 ml barium sulfate suspension 0.1% w/v was administered orally. CT examination revealed multiple tortuous venous channels throughout the wall of the jejunum (Figs. 2 & 3). These channels were not visualised in the arterial phase, and no hypertrophied arteries were noted. Multiple tortuous venous channels were also noted in the mesentery (Fig. 4). CT confirmed the presence of multiple liver haemangiomas and a gall bladder calculus (Figs. 3 & 4). The splenoportal axis and superior mesenteric vein were patent. There was no evidence of chronic liver disease, portal hypertension, splenomegaly or ascites.

A double-balloon enteroscopy was performed after MDCT, which showed multiple dilated tortuous submucosal vessels over a long segment of the jejunum (Fig. 5). The overlying mucosa was normal, without any ulceration. No active bleeding, mass, polyp or diverticula was noted. Based on the MDCT and enteroscopy findings, a diagnosis of phlebectasia was made. Surgical resection could not be performed due to the extensive jejunal involvement, and the patient was managed conservatively.

DISCUSSION

Only 3%–5% of patients with gastrointestinal bleeding have the site of bleeding located between the second portion of the duodenum and the ileocecal valve.⁽⁴⁾ The identification of the source of small bowel blood loss is often difficult due to its relative inaccessibility, length, free intraperitoneal location, vigorous contractility and the overlying loops.⁽⁴⁾ Owing to the inability to localise the bleeding site, these patients may present with prolonged chronic occult blood loss or recurrent episodes of melaena without a specific diagnosis.⁽⁴⁾ The common causes of small bowel bleeding include vascular lesions, ulcers, inflammatory bowel disease, neoplasms and Meckel's diverticulum.^(4,5) Vascular lesions are the commonest cause of small intestine bleeding, accounting for 70%–80% of the causes. Common vascular lesions of the small intestine include angiodysplasia, telangiectasia, arteriovenous malformation, haemangioma, phlebectasia (venous ectasia) and Dieulafoy's lesion.^(1,4)

Phlebectasias or venous ectasias are rare benign vascular anomalies of the gastrointestinal tract, with only a few reported cases to date.^(6–8) The causes and aetiological factors responsible for phlebectasia are not clear. These lesions are usually asymptomatic; however, occasionally, they may present with mild to massive gastrointestinal haemorrhage. Pathologically, these lesions are dilated submucosal veins with normal endothelial lining, usually



Fig. 4 Multidetector CT angiogram multiplanar reconstruction in coronal plane shows multiple venous channels in the mesentery (white arrow). Small liver haemangioma (black arrow) is seen with characteristic peripheral nodular enhancement.

with thin overlying mucosa. Endoscopically, they appear as multiple, bluish red nodules. Phlebectasia may occur in any segment of the gastrointestinal tract, including the small intestine.⁽⁶⁾ The jejunum is one of the commonly reported sites.^(6,8) These lesions may be multiple, or may involve the entire length of the small intestine. A close differential diagnosis of phlebectasia is varices, which is indistinguishable on imaging appearance. However, the diagnosis of varices is usually made in the setting of cirrhosis with portal hypertension or thrombosis of the splenoportal axis.

Determining the source of small intestine bleeding is challenging. The diagnostic accuracy of small bowel follow-through is inadequate for the evaluation of the small intestine.⁽²⁾ Although enteroclysis is capable of detecting mass lesions of the small intestine, it often fails to demonstrate flat vascular lesions, which are the commonest cause of bleeding in the small intestine. Digital subtraction angiography (DSA) is a useful diagnostic and therapeutic tool in patients who are actively bleeding.⁽²⁾ It can localise a site of bleeding in 50%–72% of patients with massive haemorrhage, but the yield decreases to 25%–50% when active bleeding has slowed down or stopped.⁽⁴⁾ Sometimes, DSA can diagnose non-bleeding lesions, such as angiodysplasias and small bowel tumours, based on the vascular pattern. However, venous bleeding is usually not identified.⁽²⁾ Since DSA is an invasive procedure, it is rarely used as

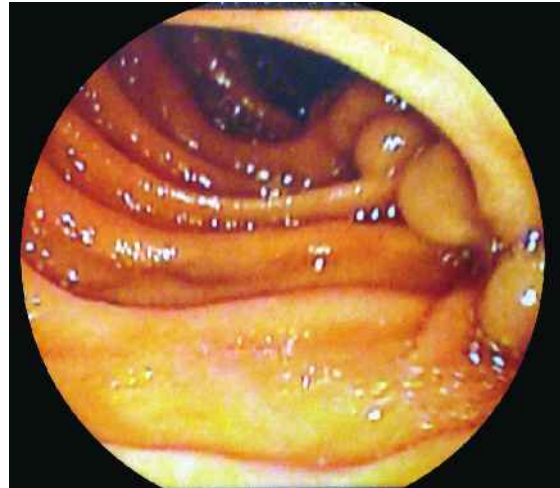


Fig. 5 Video image taken during double-balloon enteroscopy shows multiple dilated submucosal vessels in the jejunum, with normal overlying mucosa.

a first-line tool and is often performed after a positive nuclear scan. A ^{99m}Tc-labeled erythrocyte scan is a more sensitive test than DSA for the detection of minor intestinal bleeding.⁽⁴⁾ However, accurate localisation and lesion characterisation is impossible on nuclear scintigraphy, and positive scans require other modalities to localise the site of blood loss.

Double-balloon enteroscopy and capsule endoscopy are the two most important tools that ensure the evaluation of the entire small intestine.⁽⁹⁾ The recently developed double-balloon enteroscopes can be advanced much further into the small intestine than push enteroscopes. It is highly possible to evaluate the entire small intestine by double-balloon enteroscopy, using a combination of both anterograde and retrograde routes. Wireless capsule endoscopy is a new type of video telemetry system, in which a small capsule is swallowed and transmits images by a digital radio frequency communication to an external data recorder.⁽⁹⁾ The noninvasive nature of this method and its capability to visualise the entire small bowel has made it a first-line tool in many centres. The diagnostic yield of capsule endoscopy is superior to that of push enteroscopy, small bowel radiography, CT enteroclysis and mesenteric angiography in cases of obscure gastrointestinal bleeding.⁽¹⁰⁾ The limitations of capsule endoscopy include capsule entrapment and the inability to provide endoscopic-guided therapy.⁽⁹⁾ Despite these limitations, capsule endoscopy is an excellent tool for a patient who is haemodynamically stable but continues to bleed.

With advanced three-dimensional processing, MDCT angiography is now commonly used for the detection and localisation of sources of gastrointestinal

bleeding.^(5,11) In several reports on the causes of obscure gastrointestinal bleeding, unexpected bleeding foci, small intestinal tumours as well as inflammatory bowel disease have been easily detected on MDCT.⁽¹¹⁾ Vascular malformations and varices have been reported to be successfully demonstrated by MDCT.^(11,12) Similar to conventional angiography, MDCT easily visualises the bleeding points as an extravasation of contrast material in small bowel lumen.⁽¹³⁾ MDCT investigation is rapid, noninvasive, sensitive and easy to perform.⁽¹³⁾ In addition to the accurate localisation of the bleeding site, this procedure provides aetiological information in many cases. Furthermore, the concurrent visualisation of the bleeding site, extraluminal extent of the lesion and gastrointestinal vasculature is extremely helpful for interventional and surgical planning.⁽¹³⁾ However, in comparison to enteroscopy, CT is likely to miss small non-bleeding mucosal lesions and is a purely diagnostic modality.

While imaging patients with gastrointestinal bleeding, a neutral oral contrast agent should be administered, as a positive oral contrast would obscure the site of haemorrhage.^(13,14) Good luminal distension is very helpful for the delineation of small bowel lesions.⁽¹⁴⁾ A good distension of small bowel loops is usually achieved by the direct infusion of oral contrast into the small intestine with a nasojejunal tube. In a recent article, the oral administration of iso-osmotic mannitol has been described for optimum distension of the small bowel loops without the need for nasojejunal intubation.⁽¹⁵⁾

MDCT angiography has helped in identifying the location and extent of the jejunal lesions responsible for melaena in our case. Multiphasic scanning has enabled the correct characterisation of the venous nature of the jejunal lesions. Although skin and liver haemangiomas have been commonly reported in association with intestinal vascular lesions like haemangiomas and telangiectasias, they have not been reported in association with phlebectasia.⁽⁴⁾ In one case report, jejunum phlebectasia has been described with similar lesions in the scrotum and oral cavity.⁽⁷⁾ Currently, the only treatment option available for bleeding intestinal phlebectasias is surgical resection of the involved bowel.⁽⁶⁾ In view of the extensive involvement of the jejunum and the mesentery, surgery could not be performed in this case due to the concern for

short bowel syndrome.

To conclude, we present a case of jejunal phlebectasia with multiple liver haemangiomas and port-wine naevus of the anterior abdominal wall. Although enteroscopy or capsule endoscopy are the definitive tests for the evaluation of small intestine in cases of obscure gastrointestinal bleeding, our case demonstrates the potential of MDCT angiography for the detection of vascular lesions of the small intestine. MDCT angiography can be used as an alternative first-line test for the evaluation of obscure gastrointestinal bleeding as it is a rapid, noninvasive and accurate modality.

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