Deep enteroscopy diagnosis of obscure overt gastrointestinal bleeding due to intravascular endothelial hyperplasia missed by capsule endoscopy

A 56-year-old woman was referred to our hospital with a 3-day history of melena. She reported a 3-year history of dull pain in the left iliac region and intermittent melena. She had no history of drug use, fever, nausea, vomiting, or weight loss. Her vital signs were stable. She appeared pale and was tender in the left iliac region, but without guarding or rebound tenderness. Rectal examination confirmed melena. Laboratory investigation showed a red cell count of 3.16 × 10¹²/L and hemoglobin of 85.0 g/L. Other laboratory values were normal.

The results of esophagogastroduodenoscopy (EGD) and colonoscopy were negative. Capsule endoscopy also failed to identify any blood in the gastrointestinal (GI) tract. Given the possibilities of a blind spot in vision of the capsule endoscopy and intermittent bleeding of the lesion, single-balloon enteroscopy was performed when the patient presented with melena again. Enteroscopy showed a submucosal protruding lesion (approximately 0.5 × 0.5 cm) in the middle section of the jejunum that was streaming blood and had white thrombus attached to its surface. A titanium clip was therefore applied to occlude the lesion with the aim of stopping the bleeding temporarily and marking the surgical site (Fig. 1; Video 1).

Fig. 1 Single-balloon enteroscopy images showing a submucosal prominent lesion (approximately 0.5 × 0.5 cm) that was streaming with blood, with white thrombus attached to its surface: a before the titanium clip was applied; b after the titanium clip was applied.

Fig. 2 Abdominal computed tomography (CT) scan showing the location of the titanium clip (arrow) before the operation.

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Video 1

Video 1

Single-balloon enteroscopy was performed to observe intravascular papillary endothelial hyperplasia real-time in vivo. A titanium clip was applied to occlude the lesion with the aim of stopping the bleeding temporarily and marking the surgical site.

Video 1

(G Video 2). Laparoscopic partial enterectomy was performed to completely resect the lesion (Fig. 3). Postoperative histopathological examination revealed intravascular papillary endothelial hyperplasia (IPEH) (Fig. 4). The patient was doing well at follow-up 6 months later. IPEH is a benign exuberant proliferation of endothelial cells that is associated with organizing thrombus [1] and is rare in the GI tract. Here, we performed single-balloon enteroscopy and observed IPEH real-time in vivo for the first time. We found the endoscopic features of IPEH resembled an intestinal stromal tumor or hemangioma. As IPEH is a benign process, complete surgical excision, if possible, is the first-choice to cure the disease [2]. In conclusion, IPEH is rare in the GI tract. We should remain suspicious of IPEH and make the differential diagnosis from other tumors to avoid misdiagnosis or the giving of unnecessary adjuvant therapy.

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Competing interests: None
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Fig. 4 Histological appearance of the hematoxylin and eosin (H&E)-stained specimen showing an exuberant endothelial proliferation with a papillary architecture attached to the wall of a dilated vessel. The organized thrombus was entrapped by the papillae, which were covered by a single layer of flat or slightly plump endothelial cells. Original magnification: a × 20; b × 40; c × 100; d × 200.