A 26-year-old woman (case 1) was referred for evaluation of recurrent episodes of melena. Gastroscopy, colonoscopy with ileoscopy, and small-bowel computed tomography (CT) scan were normal. Capsule endoscopy (Pill cam SB 2, Given Imaging, Yoqneam, Israel) disclosed a lesion with whitish carpet-like villi and superficial red spots with spontaneous bleeding at the proximal jejunum. At double-balloon enteroscopy (Fujinon, Saitama, Japan) the lesion occupied two-thirds of the lumen (Fig. 1). The involved segment was resected by laparoscopy (Fig. 2). Microscopy showed a mixed lesion with a central core of dilated cavernous vascular channels surrounded by dilated lymph vessels (Fig. 3).

The diagnosis of a mixed cavernous hemangioma-lymphangioma was confirmed by immunostaining [1] (Fig. 4). A 59-year-old man (case 2) was admitted for two episodes of melena. Gastroscopy and colonoscopy were normal. Capsule enteroscopy revealed a polypoid lesion covered by whitish and red spots at the proximal jejunum (Fig. 5), which was confirmed on double-balloon enteroscopy.

The patient underwent single-port laparoscopy and the involved segment was resected. The lesion, 3.5 cm × 7 cm in size, corresponded to a mixed cavernous hemangioma–lymphangioma. Gastrointestinal cavernous hemangiomas are congenital benign vascular lesions that are usually located in the jejunum. Their endoscopic appearance at enteroscopy was a large hemi-circumferential lesion, with whitish carpet-like villi and red spots. The lesion was easily identified at laparoscopy because of its central bluish appearance, surrounded by whitish lymphatic tissue. The internal aspect of the surgical specimen is shown (Fig. 2). The lesion involving the mucosa and the submucosa is a cavernous hemangioma (H) surrounded by dilated cavernous lymphatic channels (L). The overlying intact mucosa is thickened by numerous lymphangiectasis (arrowhead). The asterisk indicates a focal hemorrhage in contact with the muscularis propria (hematoxylin and eosin staining; original magnification × 20).
copy or capsule endoscopy is usually of a sessile or polypoid, bluish or red lesion [2–4]. However, in our two cases, the surface of the hemangioma was covered by white spots, suggesting a lymphatic component. The mixed pattern of lymphatic-vascular tissue was confirmed on histological examination. Mixed hemangioma-lymphangioma has been previously described at the colon and the designation of hemangiolymphangioma has been proposed [5]. The images presented here are the first by means of capsule endoscopy and double-balloon enteroscopy. This histological variation should be kept in mind in the differential diagnosis of vascular lesions with lymphangiectasias.

Fig. 4 Case 1. Immunohistochemistry helped to differentiate between hemangioma and lymphangioma. Factor VIII stained the endothelium of the blood channels (left side; arrowheads), while D2–40 stained the endothelium of lymphatic channels (right side; arrows) (original magnification × 400).

Fig. 5 Case 2. Capsule enteroscopy: vascular lesion with whitish spots resembling lymphangiectasia.

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Bibliography
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