Mixed cavernous hemangioma-lymphangioma of the jejunum: detection by wireless capsule endoscopy

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 polyloid 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 includes carpet-like villi and superficial red spots.

Fig. 1 Case 1. Double-balloon endoscopy showing a large hemi-circumferential lesion, with whitish carpet-like villi and red spots.

Fig. 2 Case 1. a The lesion was easily identified at laparoscopy because of its central bluish appearance, surrounded by whitish lymphatic tissue. b Internal aspect of the surgical specimen.

Fig. 3 Case 1. 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.

Competing interests: None

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Endoscopy 2011; 43: E217–E218
© Georg Thieme Verlag KG Stuttgart · New York · ISSN 0013-726X

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Mavrogenis G et al. Mixed cavernous hemangioma-lymphangioma of the jejunum... Endoscopy 2011; 43: E217–E218