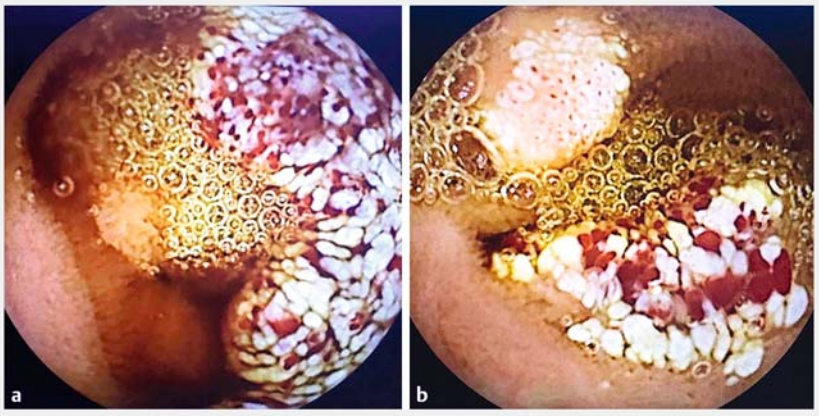
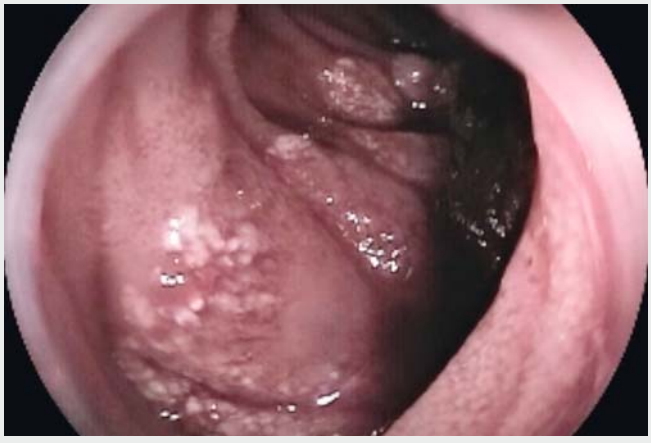
A rare cause of small-bowel bleeding: haemorrhagic small-bowel lymphangioma diagnosed by antegrade double-balloon enteroscopy



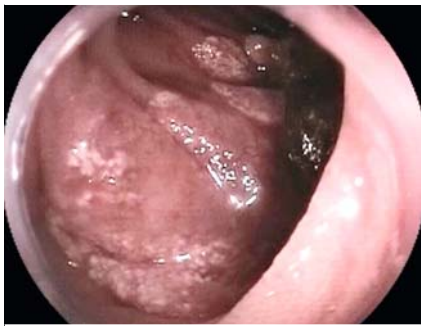
▶ **Fig. 1** Small-bowel capsule endoscopy images showing two white-speckled congested lesions, with active oozing.



Video 1 Small-bowel lymphangioma.

Small-bowel lymphangiomata (SBLs) are benign and relatively uncommon tumors of the lymphatic system [1]. Although SBLs are usually clinically silent, they may rarely present with significant small-bowel bleeding, protein-losing enteropathy, and intussusception [2, 3].

A 54-year-old-man with a past medical history of stable chronic lymphocytic leukemia presented with transfusion-dependent obscure-overt gastrointestinal (GI) bleeding. Upper and lower GI endoscopies and small-bowel cross-sectional imagining were unremarkable. A small-bowel video capsule endoscopy (VCE) showed a white-speckled congested lesion, with active oozing (▶ **Fig. 1**). The lesion was estimated to be about 2 cm in diameter and was deemed to be located within the jejunum.



▶ **Fig. 2** Image taken at double-balloon enteroscopy showing a white-yellow appearance of congested villi involved in the lesion.

Antegrade double-balloon enteroscopy (DBE) was subsequently performed. The enteroscope was advanced to an estimated insertion depth of 240 cm post-pylorus, where the lesion seen at VCE was identified (▶ **Fig. 2**, ▶ **Video 1**). This had a white–yellow appearance with overlying severely congested villi giving a "strawberry-like" mucosal pattern. The lesion was not deemed to be endoscopically resectable; multiple biopsies were taken and a reference tattoo was placed proximal to it.

Histopathological exam confirmed a lymphangiomatous etiology without any evidence of dysplasia or malignancy (▶ **Fig. 3**). Minimally invasive tattoo-guided laparoscopic resection has been planned.

This case highlights the key, complementary role of small-bowel VCE and DBE for the diagnosis and minimally invasive management of clinically significant SBLs.

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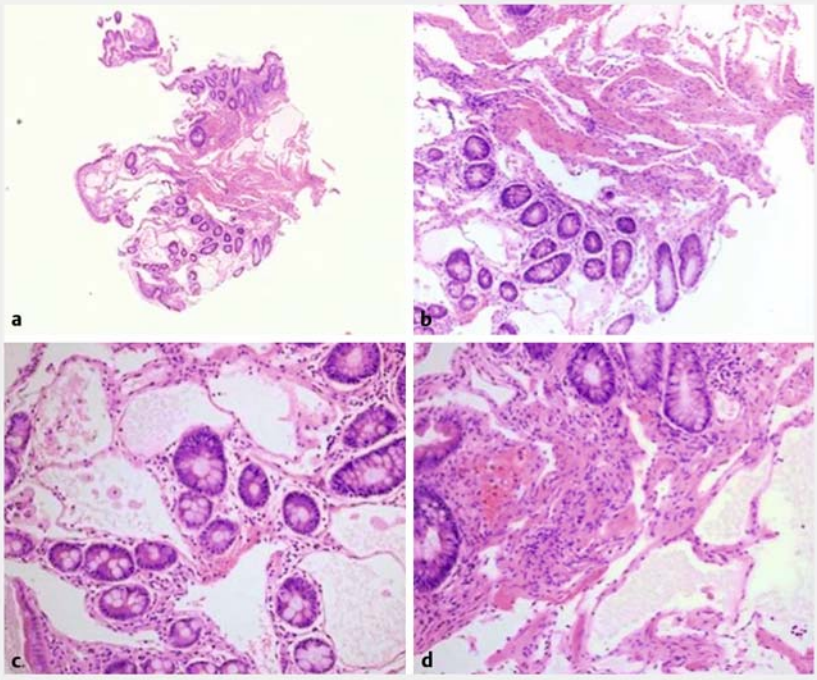
Competing interests

Dr. Despott has received research and education grants from Fujifilm, Aquilant Medical, Pentax, and Olympus.



▶ **Fig. 3** Small-bowel mucosal biopsy with hematoxylin and eosin staining captured at low magnification, showing superficial ectatic lymphatic vessel within normal villi and deeper ectatic lymphatic vessels in the submucosa. **a** ×4; **b** ×10. **c, d** Same biopsy captured at higher magnification (×20), showing superficial lymphoectasia (monolayer endothelial with no red blood cells) (**c**) and deeper ectatic lymphatic vessels beyond the muscularis mucosae (**d**).

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