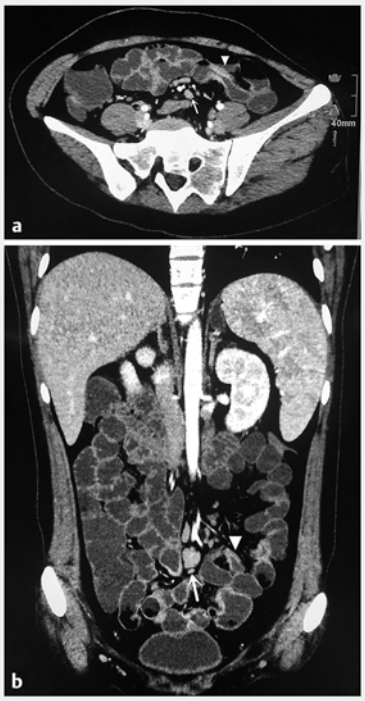
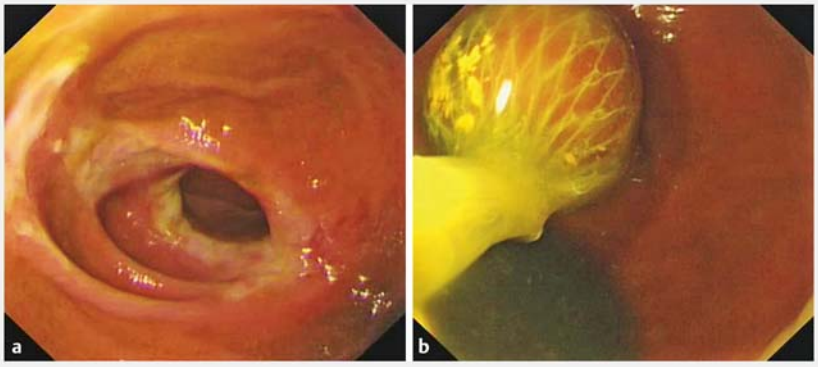
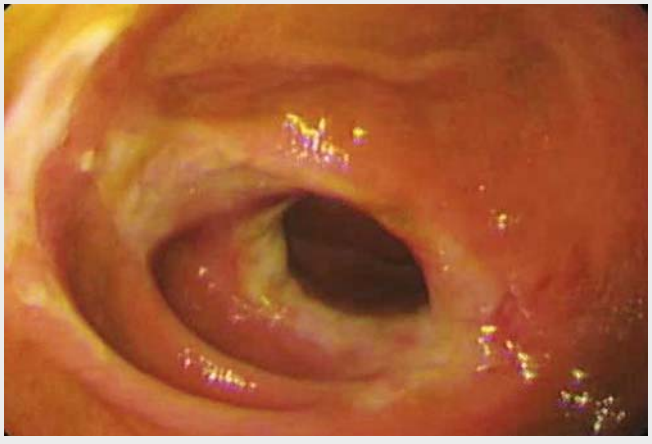
Cryptogenic multifocal ulcerous stenosing enteritis diagnosed with single-balloon enteroscopy



▶ **Fig. 1** Contrast-enhanced computed tomography enterography images showing short segmental strictures manifested within the thickened small bowel (arrowheads) and slightly enlarged mesenteric lymph nodes (arrows) in: **a** axial view; **b** coronal view.



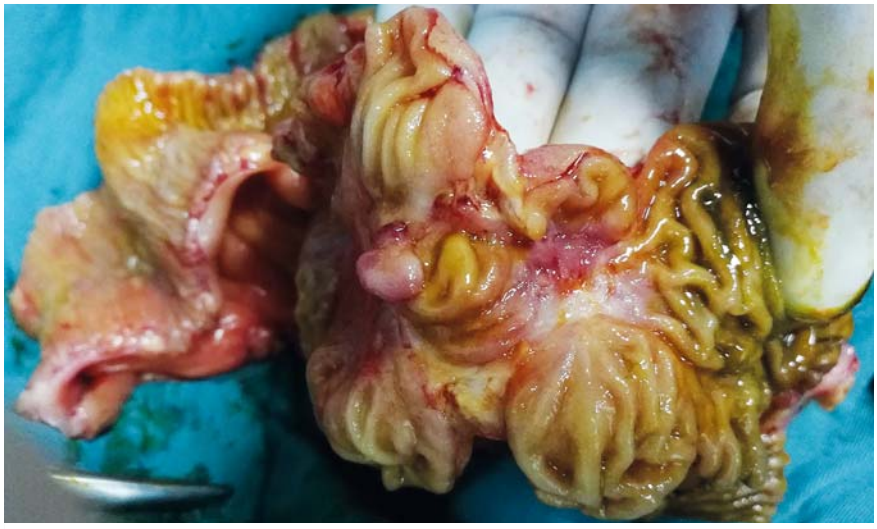
▶ **Fig. 2** Single-operator single-balloon enteroscopy views showing: **a** shallow circular ulcers covered with white moss at or near the strictures; **b** attempted removal of the retained video capsule endoscope using a net basket.



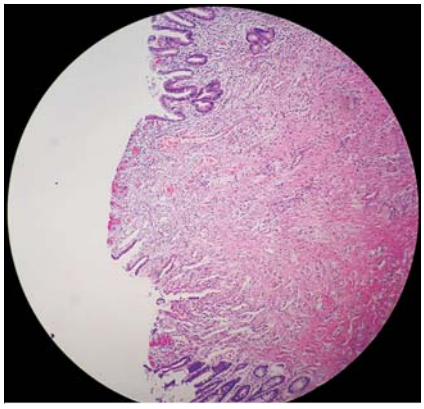
Video 1 Diagnosis of cryptogenic multifocal ulcerous stenosing enteritis (CMUSE) by single-balloon enteroscopy (SBE), with removal of a retained video capsule endoscope.

A 28-year-old woman had suffered from 6 years of chronic anemia symptoms and 4 years of repeated bouts of incomplete small-intestine obstruction with unclear cause. A retained video capsule endoscopy (VCE) 15 months previously had shown multiple circular ulcers in the small intestine. She had no history of previous medication usage, including nonsteroidal anti-inflammatory drugs (NSAIDs). Laboratory tests showed iron deficiency anemia (hemoglobin of 65 g/L), slight hypoalbuminemia (36.6 g/L), and positive occult blood in her stool, while inflammatory markers, immunologic function, and autoantibodies were all within their normal range. Tuberculosis and viral infection were also excluded. Repeated gastrointestinal (GI) endoscopy showed no remarkable findings. Computed tomography enterography (CTE) confirmed strictures manifested within the thickened small bowel and normal mesenteric vasculature (▶ **Fig. 1**).

Single-operator single-balloon enteroscopy (SBE) demonstrated multiple shallow circular ulcers with luminal narrowing in the lower ileum. The endoscopist tried to remove the retained video capsule using a net basket, but finally failed because the capsule became incarcerated in another obstructive stenosis (▶ **Fig. 2**; ▶ **Video 1**). Laparoscopy-assisted small-bowel segmental resection was finally performed (▶ **Fig. 3**). Final pathology revealed mucosal chronic inflammation and reactive hyperplasia of the enlarged



▶ **Fig. 3** Gross pathological specimen of the resected small intestine showing multiple ulcers corresponding to the single-balloon enteroscopy images, with suspected perforation found near the retained capsule.



▶ **Fig. 4** Microscopic appearance of the surgical specimen showing multiple ulcers affecting the mucosa and submucosa, hyperplasia of inflammatory granulation tissue, and nonspecific inflammatory infiltrates. No giant cell granulomas, vasculitis, or fissural ulcers were seen (hematoxylin and eosin [H&E] stain; magnification × 100).

lymph nodes located in the ileal mesentery (▶ **Fig. 4**).

We concluded clinically that her diagnosis was cryptogenic multifocal ulcerous stenosing enteritis (CMUSE), which is a rare disease characterized by repeated anemia or obstruction resulting from multiple shallow ulcers with strictures in the small intestine [1]. To our knowledge, this is the first reported CMUSE case with an enteroscopy video showing real-time observation of the characteristic circular ulcerative lesions. This case highlights that less frequent etiologies in the small bowel should also be kept in mind when dealing with chronic GI bleeding and recurrent abdominal pain, even if the symptoms are tolerable. Thus, we could treat the disorder at its non-fibrotic stage and prevent unnecessary surgery, given the fact that steroids are effective in most cases [2].

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Competing interests

None

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Bibliography
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