A forethought about obscure gastrointestinal bleeding: an unusual ileal mass

The patient was a 32-year-old woman admitted for hematochezia, with a hemoglobin of 67 g/L and fecal occult blood test of 4+. Emergency gastroscopy and colonoscopy were unremarkable. A double balloon enteroscopy revealed a lobulated ileal mass located 70 cm proximal from the ileocecal valve (▶Fig. 1, ▶Video 1). Positron emission tomography/computed tomography (PET/CT) showed that the lesion was located in the pelvic segment of the small intestine, with a standardized uptake value index of 3.6 (▶Fig. 2). Surgery was recommended owing to the patient’s low hemoglobin levels and risk of recurrent gastrointestinal bleeding. A gross specimen revealed a lobulated, polypoid mass in the ileum, measuring up to 1.5 cm × 0.8 cm (▶Fig. 3).

A final diagnosis of neuromuscular and vascular hamartoma of the small intestine was made. Postoperative pathology revealed a mixed component of blood vessels, nerve fibers, and smooth muscle consistent with that of a hamartoma (▶Fig. 4). The haphazard arrangement of the vascular structures and muscular tissues were confirmed by CD31 and desmin staining, respectively. Aberrant nerve bundles and ganglion cells were highlighted by S100 staining (▶Fig. 5).

Neuromuscular and vascular hamartoma is a rare gastrointestinal lesion first described in 1982 by Fernando and McGo-

▶Fig. 1 Endoscopic view of the lesion.

▶Video 1 Retrograde double-balloon enteroscopy was performed using the water-exchange method.

▶Fig. 2 Positron emission tomography/computed tomography revealed a lesion with increased uptake in the pelvic segment of the small intestine.

▶Fig. 3 Macroscopic view of the lesion after surgical resection.
Clinical symptoms can be non-specific and can range from chronic abdominal pain and intermittent intestinal obstruction to gastrointestinal bleeding. This condition can occur as single or multiple strictures or a polypoid mass. It is mainly composed of disorganized fascicles of blood vessels, smooth muscle, and bundles of non-myelinated nerve fibers with scattered abnormal ganglion cells, and occurs focally within a segment of the small intestine [2]. Given that similar histological features seen in cryptogenic multifocal ulcerous stenosing enteritis or diaphragm disease of the small bowel, the hamartomatous nature of neuromuscular and vascular hamartoma has been argued [3]. However, our case demonstrated a rare single, lobulated polypoid lesion of the small intestine with histological features consistent with neuromuscular and vascular hamartoma and cannot be attributed to other reactive diseases.

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Competing interests

The authors declare that they have no conflict of interest.

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References


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