

CASE REPORT

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# Multiple intestinal lymphangiomas with episodic hemorrhage requiring partial laparoscopic resection: a case report

Hiroka Kondo<sup>1\*</sup> , Takeshi Ohki<sup>1</sup>, Shimpei Ogawa<sup>1</sup>, Teppei Omori<sup>2</sup>, Hiromi Onizuka<sup>3</sup>, Yoji Nagashima<sup>3</sup> and Shigeki Yamaguchi<sup>1</sup>

## Abstract

**Background:** Lymphangioma is a non-epithelial tumor marked by aggregates of abnormally dilated lymphatics. Mesenteric occurrences account for < 1% of all cases, and < 0.05% involve the gastrointestinal tract. Most are confined to children, rarely affecting adults.

**Case presentation:** Herein, we describe an elderly Japanese woman with anemia, hypoalbuminemia, and episodic bleeding due to multiple intestinal lymphangiomas. Abdominal computed tomography revealed multiple low-density defects of mesentery, with areas of intermediate (T1 images) or high (T2 images) signal intensity similarly dispersed in magnetic resonance scanning sequences. Single-balloon enteroscopy was undertaken, enabling identification and tattooing of a small intestinal bleeding source. Laparoscopy-assisted resection at this site served to control related hemorrhage, removing a histologically confirmed hemolymphangioma. Having recovered uneventfully, the patient remained stable 2 months postoperatively.

**Conclusions:** Although rare in adults, mesenteric or gastrointestinal lymphangiomas must be considered in a setting of anemia and hypoalbuminemia. Complete resection is advantageous to improve patient symptoms, but limited resection of multiple lesions may be equally effective.

**Keywords:** Lymphangioma, Mesenteric, Intestinal, Anemia, Laparoscopic-assisted surgery

## Background

Lymphangioma is a non-epithelial growth composed of dilated lymphatic channels [1]. Mesenteric lesions are quite rare (< 1% of such tumors) [2] and may remain asymptomatic, present as distinct abdominal masses, or culminate in acute abdomen [2, 3]. Once bleeding occurs, hemostasis is largely achieved through upper gastrointestinal (GI) endoscopy or enteroscopy, reserving surgery for difficult cases [4–8]. Herein, we report an elderly woman with multiple mesenteric and intestinal lymphangiomas that bled repeatedly. The culprit site was tattooed during

small bowel enteroscopy, and laparoscopic-assisted surgical resection followed.

## Case presentation

A 77-year-old Japanese woman with chronic anemia (20+ years) had engaged elsewhere in supplementation for presumed iron deficiency. Her anemic condition seemed to abate until 6 years prior, at which point another episode prompted upper GI endoscopy and bone marrow examination. No abnormalities were apparent at the time, but fecal testing was positive for occult blood. Eventually, she was referred to our hospital in consultation. Subsequent colonoscopy and enteroscopy revealed numerous mesenteric and intestinal lymphangiomas in conjunction with bleeding. Given her fluctuating status

\*Correspondence: kondo.hiroka@twmu.ac.jp

<sup>1</sup> Department of Surgery, Institute of Gastroenterology, Tokyo Women's Medical University, 8-1, Kawada-cho, Shinjuku-ku, Tokyo 162-8666, Japan  
Full list of author information is available at the end of the article

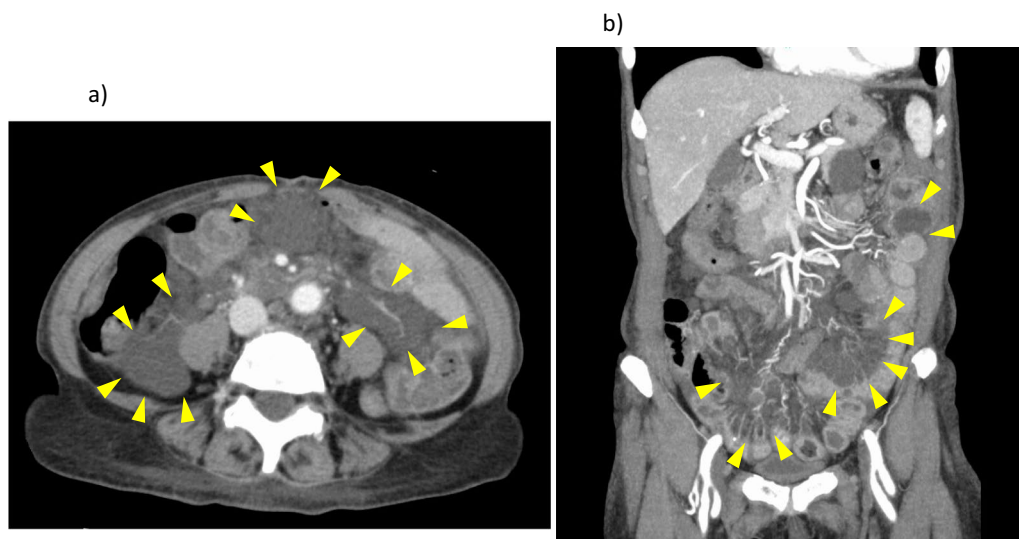
during medical treatment, often reliant on transfusions, surgical control of blood loss was the therapeutic goal.

Upon admission, the patient (height, 145 cm; weight, 50.4 kg) had a body mass index (BMI) of 24 kg/m<sup>2</sup>. Conjunctival pallor and lower leg edema were evident, but her abdomen was flat, soft, and non-tender. Other than very low hemoglobin (6.7 g/dL) and albumin (2.5 g/dL) levels, initial laboratory diagnostics were unremarkable. Her overall condition had been sustained through supportive measures, administering blood and albumin.

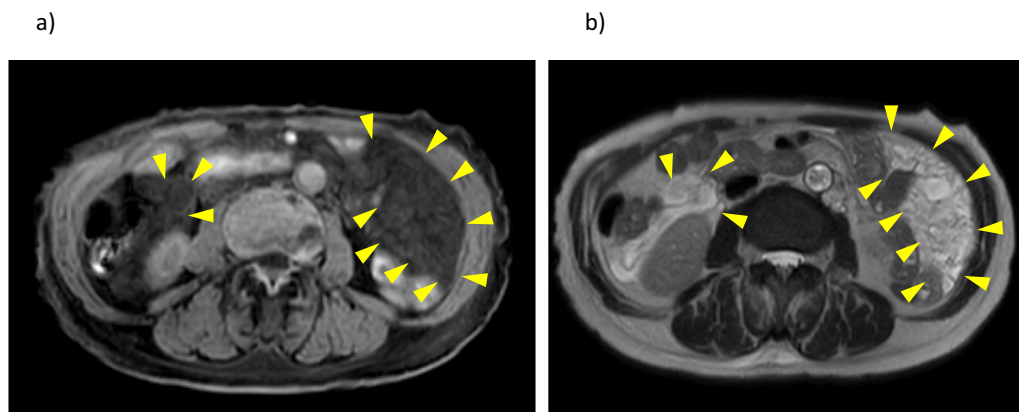
Abdominal computed tomography (CT) revealed multiple low-density mesenteric defects, devoid of contrast effect (Fig. 1). Findings of magnetic resonance imaging

(MRI) are shown in Fig. 2. There were many areas of intermediate (T1) or high (T2) signal intensity in respective scanning sequences. Capsule enteroscopy confirmed a multiplicity of lymphangiomas along jejunum and near terminal ileum (Fig. 3). Single-balloon enteroscopic tattooing was done, marking a suspected jejunal bleeding source for later operative intervention (Fig. 4).

Hemostasis was ultimately achieved through laparoscopic-assisted surgery. A 4-cm umbilical incision was made, installing a multi-flap gate for converter attachment and 12-mm port insertion. CO<sub>2</sub> insufflation (10 mmHg) produced a pneumoperitoneum sufficient for viewing the abdominal cavity. Many yellowish, cystic



**Fig. 1** Enhanced abdominal computed tomography: Multiple low-density areas of mesentery in **a** axial and **b** coronal views, devoid of contrast effect (arrowhead)



**Fig. 2** Abdominal magnetic resonance imaging study: Multiple lesions of **a** T1-weighted and **b** T2-weighted sequences, showing respective signal intensities (T1, intermediate; T2, high) (arrowhead)



**Fig. 3** Capsule enteroscopic delineation of multiple lymphangiomas at jejunum and terminal ileum: the surface was yellowish-white with reddish color and finely granular (arrow)

growths and hemangiomatous nodules studded the small intestine and mesentery, accompanied by low-volume, grayish-white, and slightly cloudy ascites (Fig. 5). Extracorporeal operation proceeded, once the above concluded. Having inspected and palpated the entire small intestine up to ligament of Treitz, it appeared that the preponderance of cystic mesenteric and intestinal lesions were jejunal. Their sheer number prohibited complete resection, but partial excision was still feasible. We thus removed intestine and adjacent mesentery at the point of tattoo (to control bleeding), performing functional

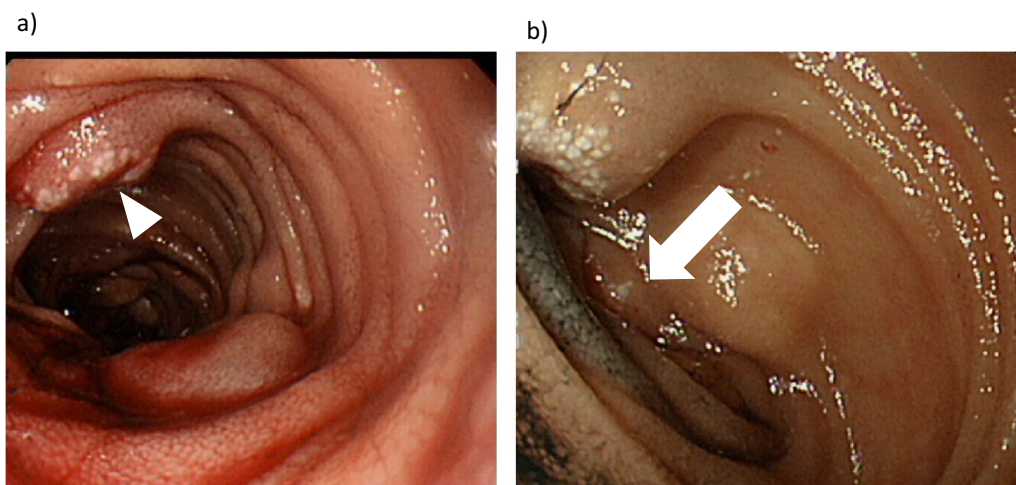
end-to-end anastomosis. Operative time was 114 min, blood loss was 3 ml, and no blood was transfused during or after the operation.

The anemia stabilized postoperatively. In Fig. 6, multiple yellowish-white submucosal lesions are grossly visible, seen as a profusion of dilated lymphatic vessels in hematoxylin and eosin-stained histologic sections. These lymphoid structures were proven to be positive for podoplanin by immunostaining, and negative for CD31. The patient recuperated without incident and was discharged 8 days after surgery. At postoperative Month 2, no worsening anemia or hyperalbuminemia had developed.

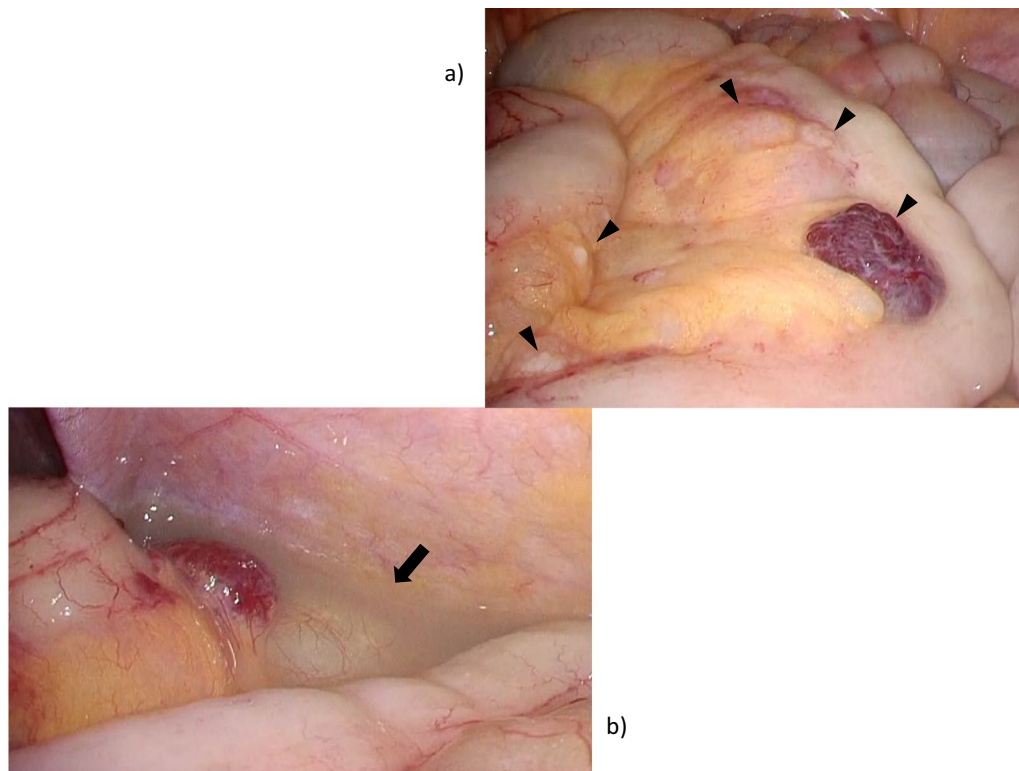
### Conclusions and discussion

Mesenteric lymphangiomas are rare congenital malformations of the lymphatic system, presenting as non-epithelial, neoplastic proliferations of dilated lymphatic channels [1]. The most common sites of origin are head and neck region or axilla, together contributing >95% of cases [9]. GI lesions account for <0.05% [10]. Definitive diagnosis relies on histologic features, using depth and size of abnormal lymphatics to recognize four tumor subtypes as follows: capillary, cavernous, cystic (hygroma), or hemolymphangioma (mixed vascular/lymphatic elements) [11]. Lesions in this patient corresponded with the latter. According to past reports, lymphangiomas resected for hemostasis have been conspicuous growths [4, 6, 12–14], harboring putative vascular constituents that promote blood loss.

Most mesenteric lymphangiomas (65%) are discovered at birth, and nearly all (90%) are reportedly diagnosed by age 2 [15]. Lymphangiomas of small intestine rarely occur, comprising 1–2% of benign small bowel tumors [16]. Without benefit of enteroscopy, they defy



**Fig. 4** **a** Enteroscopy revealed an area that appeared to be bleeding heavily (arrowhead). **b** Enteroscopic tattoo applied to the same area (arrow)



**Fig. 5** Operative findings: **a** array of yellowish, cystic lesions and hemangiomatous tumors dispersed along small intestine and mesentery (arrowhead) and **b** small volume of grayish-white, slightly cloudy ascites (arrow)

preoperative detection [17]. This case is fairly emblematic, failing to amass conclusive evidence despite long-standing anemia and repeated diagnostic testing. Enteroscopy is thus a worthwhile pursuit in any patient with obscure yet progressive anemia and hypoalbuminemia, bearing lymphangioma in mind.

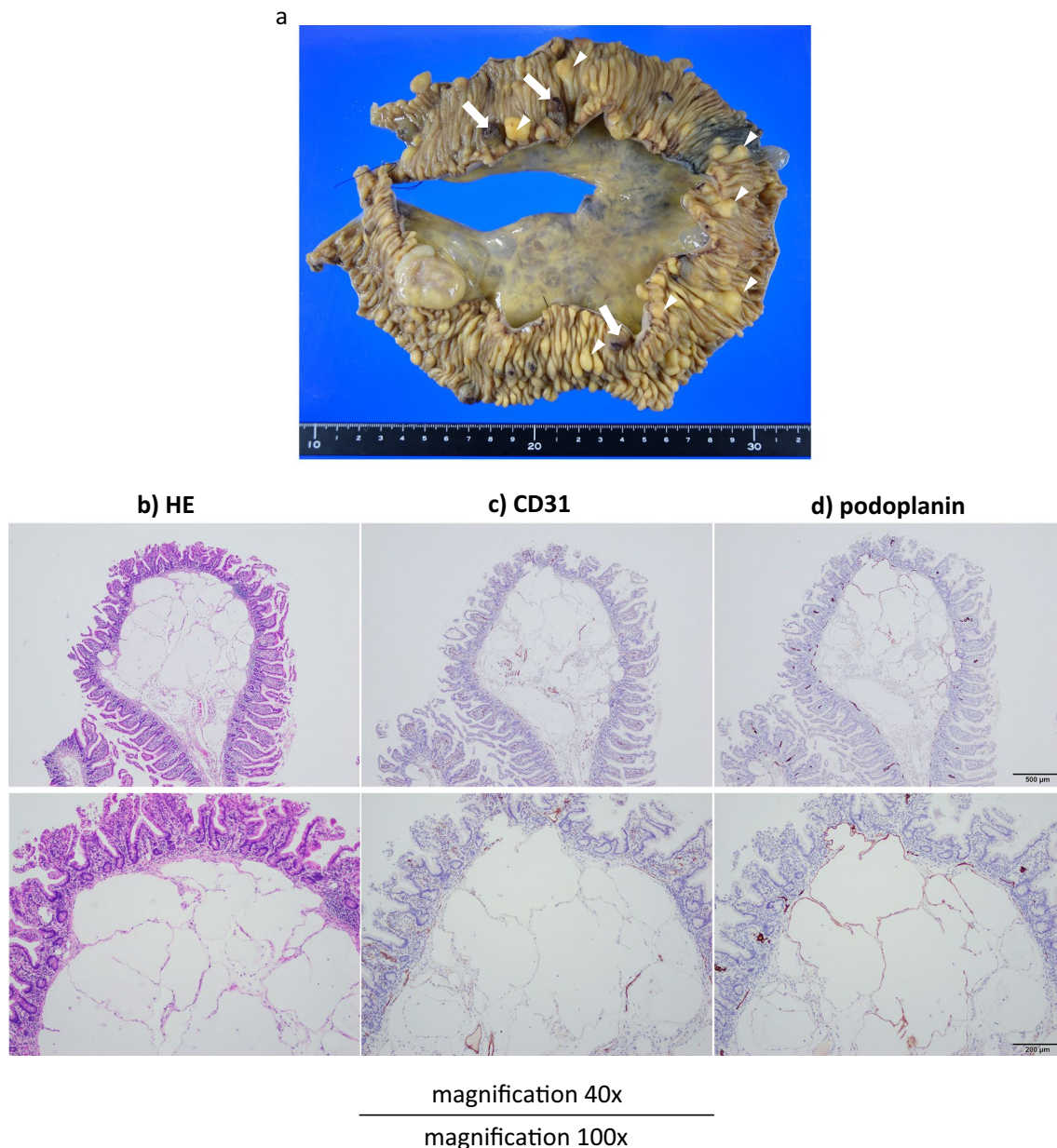
While generally benign in nature, such tumors may pervade mesenteric roots and peripheries (at intestinal wall) or invade surrounding organs, with potentially life-threatening complications. Traumatic rupture (causing intra-abdominal or intracavitary bleeding and anemia), ischemic tissue necrosis, intestinal gangrene (due to axial torsion), and intermittent intestinal obstruction are possible [18, 19]. GI bleeding was episodic in our patient and difficult to control by medical means. The alternative was surgical resection. However, other published reports [4, 6, 8, 20–22] have warned of the elusiveness posed by bleeding sites amidst multiple lesions.

Ugwonali et al. have used intraoperative enteroscopy to identify and excise sources of hemorrhage in this setting, achieving good results [23]. Morris-Stiff et al. have also installed preliminary tattoos during small bowel endoscopy as flags for resection [7]. Our patient had numerous growths, often adjacent to intestinal wall, limiting the

traceability of active bleeding by intraoperative palpation alone. Enteroscopy performed in advance of surgery to pinpoint and tattoo a suspected bleeding site helped expedite intraoperative resection of the culprit segment.

Complete resection of a localized lymphangioma is of course ideal [19, 24]. However, partial resection constitutes a valid approach to multiple lesions, offering substantial volume reduction and serving to improve patient quality of life [25]. Residual neoplastic involvement is a lingering concern that must be addressed through careful follow-up monitoring.

Herein, we describe a patient with multiple mesenteric and intestinal lymphangiomas who experienced chronic episodic bleeding. The situation was resolved through enteroscopic tattoo and laparoscopic-assisted surgical resection. In adults, mesenteric and GI lymphangiomas are rare occurrences to bear in mind as underlying causes of anemia and hypoalbuminemia. Although complete resection is best for symptom improvement, partial resection of identified bleeding points may likewise enhance patient quality of life. Preliminary enteroscopic tattooing is a means of facilitating minimally invasive surgery in these circumstances.



**Fig. 6** Gross and microscopic tumor presentation: **a** multiple yellowish-white submucosal lesions (arrowhead on typical ones), and some with traces of post-hemorrhage (arrow); **b** hematoxylin and eosin stain, magnification  $\times 40$  and  $\times 100$ ; **c** CD31 immunostain, magnification  $\times 40$  and  $\times 100$ ; and **d** podoplanin immunostain, magnification  $\times 40$  and  $\times 100$ . These lymphoid structures were proven to be positive for podoplanin by immunostaining, and negative for CD31

#### Abbreviations

BMI: Body mass index; CT: Computed tomography; GI: Gastrointestinal; MRI: Magnetic resonance imaging.

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#### Authors' contributions

HK drafted the manuscript, TO directed the preoperative investigation, and SY provided academic advice. HK and TO jointly performed the operation. YN and

HO conducted pathologic evaluations. All authors read and approved the final manuscript.

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#### Availability of data and materials

All data generated or analyzed during this investigation are included in the published manuscript.

## Declarations

### Ethics approval and consent to participate

The Institutional Review Board at Tokyo Women's Medical University has stipulated that ethical approval is unnecessary for case reports. Consent was obtained from the sole patient participant.

### Consent for publication

The patient granted consent for publication of this report.

### Competing interests

The authors have no competing interests to declare.

### Author details

<sup>1</sup>Department of Surgery, Institute of Gastroenterology, Tokyo Women's Medical University, 8-1, Kawada-cho, Shinjuku-ku, Tokyo 162-8666, Japan. <sup>2</sup>Department of Gastroenterology, Tokyo Women's Medical University, 8-1, Kawada-cho, Shinjuku-ku, Tokyo, Japan. <sup>3</sup>Department of Surgical Pathology, Tokyo Women's Medical University, 8-1, Kawada-cho, Shinjuku-ku, Tokyo, Japan.

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