Blue rubber bleb nevus syndrome (BRBNS) is a rare congenital disease with venous malformations on the skin and in the gastrointestinal tract. Gastrointestinal venous malformations frequently cause bleeding and/or iron deficiency anemia [1]. Endoscopic approaches such as endoscopic mucosal resection [2], electrocoagulation, sclerotherapy, and ligation [3] have been reported for the treatment of symptomatic gastrointestinal hemangiomas associated with BRBNS. Recently, underwater endoscopic mucosal resection (UEMR) has rapidly been becoming a game-changing technique for endoscopic polyp resection. UEMR is usually simpler, cheaper, and more reliable than other conventional endoscopic resection techniques. When endoscopic ultrasonography (EUS) is performed prior to UEMR, EUS can allow prediction of the safety and reliability of UEMR because lesion characteristics such as depth, blood vessels, and echo-density are evaluated [4]. We illustrate a case in which colon hemangiomas compatible with BRBNS were resected endoscopically using UEMR.
A 35-year-old man was referred for evaluation of a blue polyp in the cecum and another in the transverse colon found on colonoscopy after a positive fecal immunochemical test. The patient had undergone surgical resection of skin hemangiomas on his right leg at 1 and 4 years of age. Outpatient colonoscopy in our institution revealed elevated blue lesions, one in the cecum and one in the transverse colon. Magnifying narrowband light examination (EC-760ZP-W/M, Fujifilm, Tokyo, Japan) with a distal attachment (D-201-14304, Olympus, Tokyo, Japan) using blue-light imaging did not show the typical vascular pattern of a neoplasm but showed a normal surface pattern. The characteristics were classified as type 1 (Japan NBI Expert Team classification), with normal mucosa (Fig. 1). EUS (EUS-ME1, Olympus) clearly demonstrated an isoechoic/slightly hyperechoic mass in the submucosa without dilated blood vessels and the submucosal layer between the mass and the muscularis (Fig. 2). The cecal lesion had similar EUS characteristics. With cecal contraction, the lesions became floating subpedunculated tumors on the underwater endoscopic view (Fig. 3a). This suggested that complete endoscopic resection using UEMR was both safe and feasible on an outpatient basis. For both lesions, the tip of the snare (15-mm Rota Snare, Medi-Globe GmbH, Achenmühle, Germany) was securely placed on normal mucosa beyond the lesion with a sufficient proximal margin. The snare was gradually closed, always ensuring that the snare captured the entire lesion with its surrounding normal mucosa, while at the same time the water was aspirated. Once the snare was closed, the lesion was completely captured inside it. The secured lesion was cut with coagulation-mode diathermy (ESG-100, Olympus). The hemangioma was removed endoscopically. The mucosal defect was closed with a re-openable clip (Sureclip Plus, Micro-Tech Co. Ltd., Nanjing, China) and endoclips (EZ-clip, Olympus). For both lesions, UEMR was completed without complications (Fig. 3b). Pathologic evaluation revealed submucosal hemangiomas compatible with BRBNS (Fig. 4).

This patient’s case demonstrates that colon hemangiomas compatible with BRBNS can be safely and completely resected using UEMR after the submucosal characteristics have been confirmed using EUS.

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Competing interests

H. Yamamoto has a consultant relationship with the Fujifilm Corporation and has received honoraria, grants, and royalties from the company. The other authors have no conflicts of interest to disclose.

The authors

Yuko Shibuya1, Masahiro Okada1, Munefumi Arita2, Yoshikazu Hayashi1, Hirotoshi Kawata3, Alan Kawarai Lefor2, Hironori Yamamoto1

1 Department of Medicine, Division of Gastroenterology, Jichi Medical University, Shimotsuke, Japan
2 Department of Surgery, Jichi Medical University, Shimotsuke, Japan
3 Department of Diagnostic Pathology, Jichi Medical University, Shimotsuke, Japan

Corresponding author

Yoshikazu Hayashi, MD, PhD
Department of Medicine, Division of Gastroenterology, Jichi Medical University, 3311-1 Yakuji, Shimotsuke, Tochigi, Japan
hayashi@jichi.ac.jp

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