Cyanoacrylate glue in the management of blue rubber bleb nevus syndrome

Case 1. A 16-year-old boy with blue rubber bleb nevus syndrome (BRBNS) was admitted for occult bleeding. He had a history of gastric and colonic hemangiomas treated 2 years earlier by argon plasma coagulation and alcohol injection respectively. Clinical examination showed a few hemangiomas on his back and feet (Fig. 1).

Capsule endoscopy (Pillcam SB2; Given Imaging, Yoqneam, Israel) disclosed three blebs of the small bowel (Fig. 2), and the patient underwent double-balloon enteroscopy (Fujinon, Saitama, Japan). A 2-cm bleb was found at the jejunum, two smaller lesions at the ileum, and one at the transverse colon. N-butyl-2-cyanoacrylate and methacryloxyisulfolane (Glubran 2), 1 ml, diluted with Lipiodol, 1 ml, was injected in aliquots of 1 ml per injection and up to 4 ml at each lesion (Fig. 3). No complications occurred. At 6-month follow-up the patient had no anemia.

Case 2. An 11-year-old girl with BRBNS was addressed for hematochezia due to colonic blebs. Colonoscopy revealed a bleeding lesion at the cecum (Fig. 4). After initial failure to stop the bleeding with a hemostatic grasper (Coagrasper, Olympus), Glubran 2 was injected successfully. Two more blebs of the right colon were treated similarly. After 2 years the patient remained asymptomatic.
BRBNS is a rare condition characterized by multiorgan cavernous hemangiomas. Gastrointestinal bleeding is the major problem, and several endoscopic therapeutic approaches have been proposed: Nd:YAG laser, bipolar or argon plasma coagulation, band ligation, snare resection, and sclerotherapy [1–4]. Our patients were treated successfully by Glubran 2 injection, commonly used in the management of gastric varices. Endoscopic obliteration of cavernous hemangiomas with n-butyl-2-cyanoacrylate (Histoacryl) has been previously described, in the rectum [5]. However, these appear to be the first cases of cyanoacrylate glue injection to control bleeding in the small bowel and colon in the setting of BRBNS.

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References

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