Blue rubber bleb nevus syndrome: endoscopic treatment with sclerotherapy during double-balloon enteroscopy in a 9-year-old boy

Blue rubber bleb nevus syndrome (BRBNS), also called Bean syndrome, is a rare disease associated with multiple venous malformations essentially in the skin and the gastrointestinal (GI) tract [1, 2]. It commonly presents with anemia and patients respond to supportive measures, but severe symptoms may be treated by surgical resection, endoscopic sclerotherapy, and laser photocoagulation [3, 4].

We report here a case of BRBNS presenting with GI bleeding in a 9-year-old boy who was first diagnosed at the age of 1, with a venous angioma in the knee. The onset of melena led to a complete exploration of the upper and lower GI tract, and capsule endoscopy, which revealed several typical blue lesions in the stomach (▶Fig. 1a, b), small bowel (▶Fig. 2), and colon (▶Fig. 1c).

Gastric and colonic lesions were first treated with argon plasma coagulation (▶Fig. 3) in the pediatric hospital. The patient was then referred to our unit for double-balloon enteroscopy (EN-580T; Fujifilm, Tokyo, Japan), through which we could identify five pedunculated lesions and four flat friable ones. The progression of the endoscope was difficult owing to the small size of the intestinal loops; however, successful sclerotherapy of the lesions was achieved by injecting aetoxisclerol (▶Video 1). Apart from these angiomas, no further lesions required treatment, according to capsule endoscopy. The patient did not experience any new bleeding after this treatment.

In conclusion, BRBNS is rare but the diagnosis has to be considered when typical lesions are seen in different areas. Double-balloon enteroscopy using an adult enteroscope appears to be feasible in children, but progression is probably more difficult in a small-diameter bowel. As previously demonstrated, aetoxisclerol seems to be effective for the treatment of lesions with low risk of perforation.

Endoscopy_UCTN_Code_TTT_1AP_2AD

E-Videos

▶Fig. 1 Endoscopic view of the angiomas. a Gastric antral lesion. b Gastric pre-pyloric lesion. c Colonic lesion.

▶Video 1 Video capsule endoscopy diagnosis and double-balloon endoscopic sclerotherapy of gastrointestinal angiomas.

Competing interests

None
Fig. 2 Capsule endoscopic views of angiomas in the small bowel.

Fig. 3 The lesions after treatment with argon plasma coagulation. a Gastric antral lesion. b Gastric pre-pyloric lesion. c Colonic lesion.
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References


Bibliography

DOI https://doi.org/10.1055/a-0840-3214
Published online: 2019
Endoscopy
© Georg Thieme Verlag KG
Stuttgart · New York
ISSN 0013-726X