

Blue rubber bleb nevus syndrome: an unusual cause of intestinal bleeding in the elderly



► **Fig. 1** Capsule endoscopy shows many blue angiodysplasias along the jejunum.

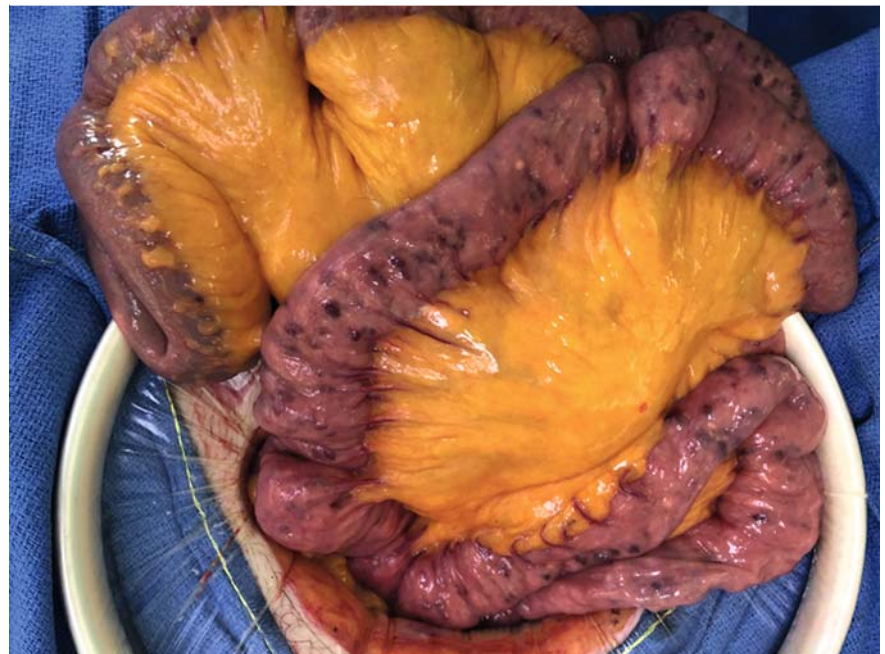


► **Video 1** Diagnosis and management of a small-bowel blue rubber bleb nevus syndrome.

Blue rubber bleb nevus syndrome (or Bean syndrome) is a rare cause of gastrointestinal bleeding. Only three cases have been reported in the elderly [1–3].

A 70-year-old man was hospitalized for recurrent and severe digestive bleeding for one month. He had recently undergone coronary stent surgery requiring antiplatelet and anticoagulant treatments, leading to lower gastrointestinal (GI) bleeding and severe anemia. Daily blood transfusions were required. Oesogastro-duodenal endoscopy and colonoscopy were normal. A small bowel capsule endoscopy showed many ectasia of the vessels along the small bowel with active and diffuse bleeding, suggesting blue rubber bleb nevus syndrome (► **Fig. 1**, ► **Video 1**). Abdominal computed tomography (CT) scan and multiphase CT enterography showed arteriovenous malformations in the jejunum and ileum without active bleeding. However, they were too numerous to be treated by endoscopic coagulation. Radiological embolization was not possible because of the serious risk of mesenteric ischemia. We decided to perform an exploratory laparotomy in conjunction with an intraoperative endoscopy of the small bowel.

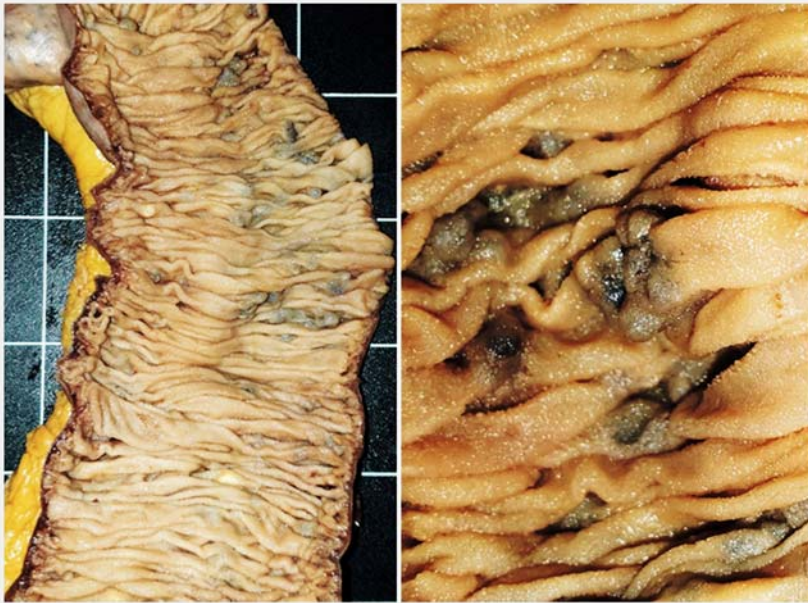
We observed many transperietal angiodysplasias along 3 meters of the small



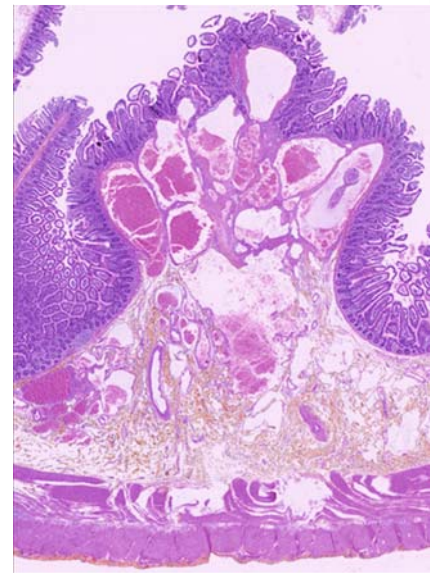
► **Fig. 2** Exploratory laparotomy shows many transperietal angiodysplasias along the small bowel. Most of them are blue, but some are yellowish.

bowel starting at the angle of Treitz (► **Fig. 2**). The ileum seemed to be free of these lesions. The intraoperative enteroscopy confirmed that there were no le-

sions in the ileum and 100 arteriovenous malformations in the jejunum with some active bleeding. We decided to perform an extensive bowel resection, removing



► **Fig. 3** Macroscopic view shows flat and polypoid submucosal lesions. Most of them are blue, but some are yellowish.



► **Fig. 4** Microscopic analysis of benign angiodysplasias with hemangiomatous and lymphangiomatous components.

the entire jejunum and keeping about 180 cm of ileum, with a handsewn end-to-end anastomosis between the proximal jejunum and the ileum. The patient was discharged on postoperative day 12. The pathological analysis identified flat and polypoid blue and yellowish lesions in the submucosa (► **Fig. 3**). The microscopic aspect revealed hemangiomatous and lymphangiomatous components (► **Fig. 4**). Six months later, the patient was well, with stable hemoglobin and no recurrent GI bleeding.

This case demonstrates the need for a combined endoscopic and surgical approach to treat extensive jejunal blue rubber bleb nevus syndrome.

Endoscopy_UCTN_Code_CCL_1AC_2AB

Competing interests

The authors declare that they have no conflict of interest.

The authors

Jérôme Winkler¹, Marion Bonnet², Aurélie Haffner³, Géraldine Gascou-Tessonier¹, Laurent Heyries¹, Philippe Grandval¹, Diane Mege²

- 1 Department of Gastroenterology, Aix Marseille Université, APHM, Timone University Hospital, Marseille, France
- 2 Department of Digestive Surgery, Aix Marseille Université, APHM, Timone University Hospital, Marseille, France
- 3 Department of Pathology, Aix Marseille Université, APHM, Timone University Hospital, Marseille, France

Corresponding author

Diane Mege, MD, PhD

Aix Marseille Université, APHM, Department of Digestive Surgery, Timone University Hospital, 264 Rue Saint-Pierre, 13005 Marseille, France
 Fax: +33-4-91-385355
 dr.dianemege@gmail.com

References

- [1] Aron J et al. An unusual cause of gastrointestinal bleeding in a haemodialysis patient. *Hemodial Int* 2018; 22: E60–E62
- [2] Gião Antunes AS et al. Blue rubber bleb nevus syndrome: a delayed diagnosis. *GE Port J Gastroenterol* 2017; 24: 101–103
- [3] Baker AL et al. Gastrointestinal bleeding due to blue rubber bleb nevus syndrome. A case diagnosed by angiography. *Gastroenterology* 1971; 61: 530–534

Bibliography

Endoscopy 2022; 54: E53–E54

DOI 10.1055/a-1381-6267

ISSN 0013-726X

published online 5.3.2021

© 2021. Thieme. All rights reserved.

Georg Thieme Verlag KG, Rüdigerstraße 14, 70469 Stuttgart, Germany