

Title:

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Obscure gastrointestinal bleeding in the setting of blue rubber bleb nevus syndrome with extensive small bowel involvement

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Blue rubber bleb nevus syndrome (BRBNS) is a rare disorder characterized by multiple vascular malformations of the gastrointestinal (GI) tract, skin and less frequently in solid organs (1). An 85-year-old male was admitted to the ER due to melena. The patient was under apixaban treatment due to a recent ischemic stroke. Dorsal hemangiomas were identified on physical examination (Fig. 1A). On admission, the hemoglobin level was 7.6 g/dl, esophagogastroduodenoscopy was negative and colonoscopy revealed blood clots in all segments, including the terminal ileum. Capsule endoscopy revealed multiple polypoid vinaceous-colored formations in the proximal jejunum (Fig. 1B) and distally active bleeding resulting in limited mucosal observation. Abdominal computed tomography (CT) angiography did not identify the bleeding source.

Balloon-assisted enteroscopy (BAE) allowed the identification of multiple hemangioma-like purplish blue lesions in the jejunum and ileum, without active bleeding (Fig. 2). A diagnosis of BRBNS was made based on clinical, imaging and endoscopic findings. Supportive treatment was decided considering the extent of the lesions and the comorbidities of the patient.

Discussion

Treatment depends on the site, size and number of lesions. Surgical resection is more suitable for limited or life-threatening lesions. Endoscopic treatment with polidocanol, coagulation, band ligation and endoscopic mucosal resection are also available (2,3). Sirolimus has been successfully used. However, tolerability and adverse effects limit its use as a rescue therapy (2).

Author's contribution: João Carlos Silva: analysis and interpretation of the case and drafting of the article. Balloon-assisted enteroscopy execution. Edgar Afecto: capsule endoscopy examination. Hospital admission and follow-up. Adélia Rodrigues: hospital admission, diagnostic approach and therapeutic management. Endoscopic procedures execution. Rolando Pinho: revised the paper critically for important intellectual content.

Acknowledgements: Silva JC wrote the manuscript and performed balloon-assisted enteroscopy. Afecto E and Rodrigues A had a decisive role in hospital admission, endoscopic study and patient follow-up. Pinho R critically revised the paper for important intellectual content. All authors approved the final version of the manuscript.

Informed consent: Informed consent was obtained from the patient.

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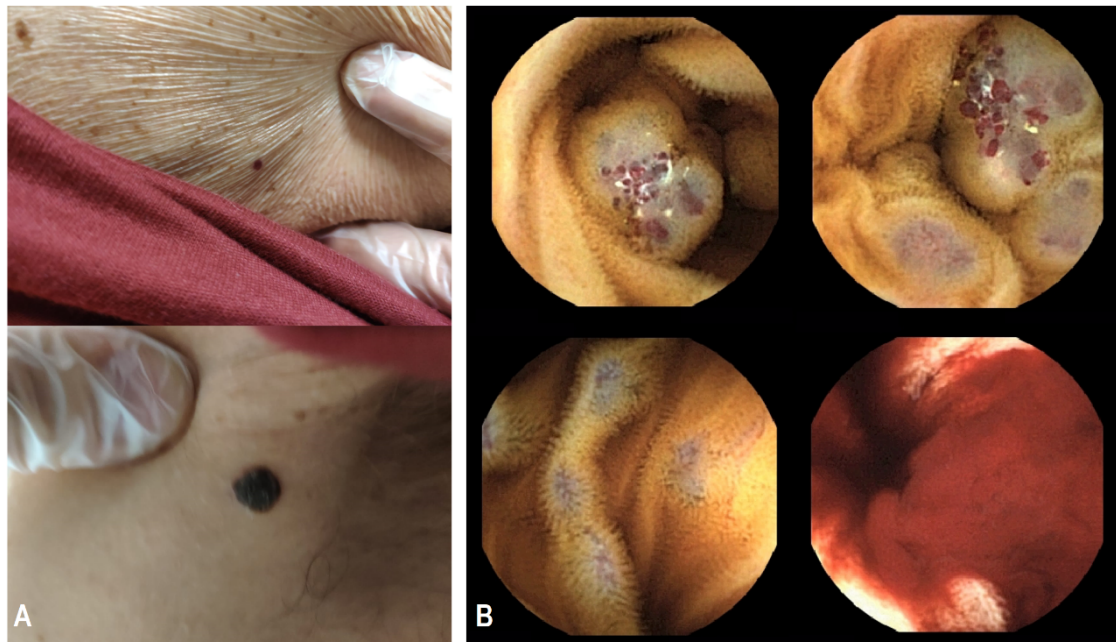


Fig. 1. Blue rubber bleb nevus syndrome. A. Physical examination identified bluish nodular cutaneous lesions in the dorsal region. B. Capsule endoscopy revealed multiple, bluish, nodular hemangiomas in the proximal jejunum.

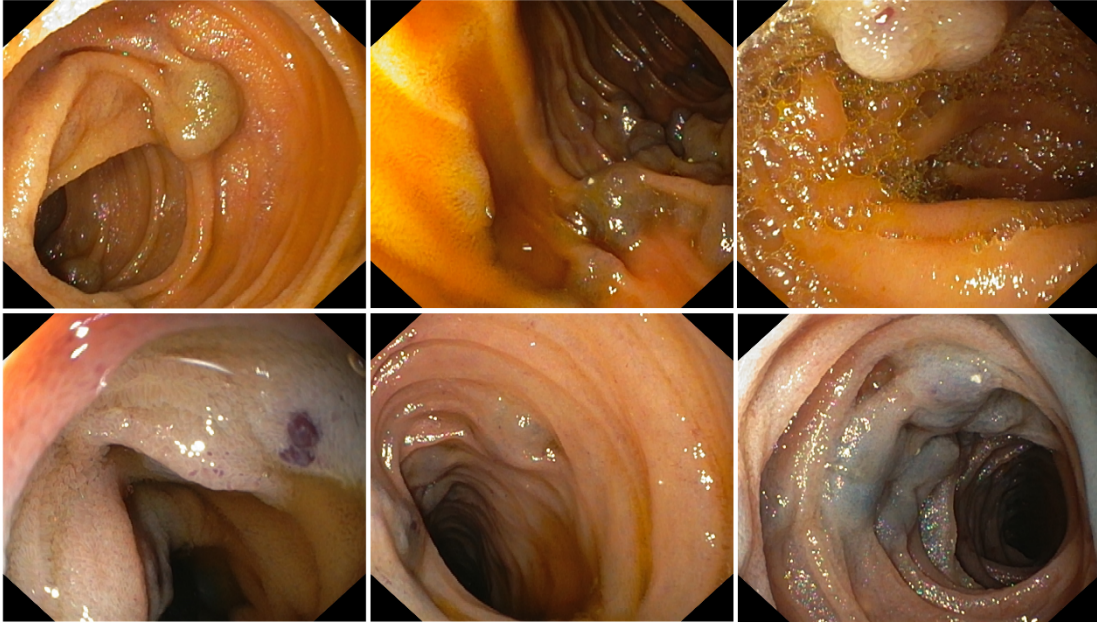


Fig. 2. Balloon-assisted enteroscopy (BAE) allowed the identification of multiple 8-30 mm hemangioma-like purplish blue lesions in the jejunum and ileum, forming conglomerates in some segments without active bleeding.