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Combined use of band ligation and detachable snares (endoloop) in a patient with blue rubber bleb nevus syndrome

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Abstract

A 20-year-old woman was admitted with iron deficiency anemia and history of melena. Upper endoscopy demonstrated normal esophagus, stomach and proximal duodenum. At colonoscopy, two bluish pedunculated angiectatic polypoid lesions (1 to 2 cm in diameter) were noted due to blue rubber bleb nevus syndrome (BRBNS). The small intestine was screened for further lesions with the use of capsule endoscopy. The capsule identified four more lesions at the proximal jejunum. The small bowel lesions were smaller, 0.5 to 1cm in diameter at their bases. We performed enteroscopy to approach the jejunal lesions. All lesions were ligated in one session using the band ligation technique with multi-band ligator device. A colonoscopy was performed and detachable snares - endoloops were inserted at the colonic lesions. Three weeks following the interventions further endoscopic evaluations were performed; they revealed scars at all sites treated with either technique. No bleeding or new venous malformations were observed. In conclusion, we report a dual technique resection method on a patient with BRBNS. The choice of resection technique depended on the lesion characteristics. Both band ligation and endoloop can be safely used for polypoid BRBNS lesions at the jejunum and colon respectively.

Keywords Blue rubber bleb nevus syndrome, band ligation, endoloop

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Introduction

Blue rubber bleb nevus syndrome (BRBNS) is a rare disease associated with multiple venous malformations in the skin, gastrointestinal (GI) tract and other internal organs. The skin lesions were first described by Gascoyen in 1860 [1,2]. It usually presents with extensive hemorrhage or iron deficiency anemia [3]. Although an autosomal dominant inheritance has been described, the majority of the cases appear to be sporadic [4].

A variety of therapeutic strategies have been proposed for the management of GI bleeding in BRBNS [5-11]. These include non-interventional (iron supplementation and/or blood transfusion, antiangiogenic agents, hormonal therapy) and interventional (endoscopic as well as surgical) measures. Endoscopic techniques reported include snare polypectomy, band ligation, YAG laser, bipolar, Argon plasma electrocoagulation, sclerosis and laser photocoagulation.

This case report aims to describe the management of BRBNS with the combined use of band ligation in the jejunum and endoloop in the colon, a technique not previously described.

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Conflict of Interest: None

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Case report

A 20-year-old woman was admitted with iron deficiency anemia and history of melena. Upper endoscopy demonstrated normal esophagus, stomach and proximal duodenum. At colonoscopy, two bluish pedunculated angiectatic polypoid lesions (1 to 2 cm in diameter) were noted due to BRBNS (Fig. 1A). The small intestine was screened for further lesions with the use of capsule endoscopy. The capsule identified four more lesions at the proximal jejunum. The small bowel lesions were smaller (0.5 to 1 cm in diameter at their bases). She had a similar venous malformation at the gingival area which was embolized. CT scan of abdomen, chest and brain did not reveal any other lesion.

We performed enteroscopy to approach the jejunal lesions

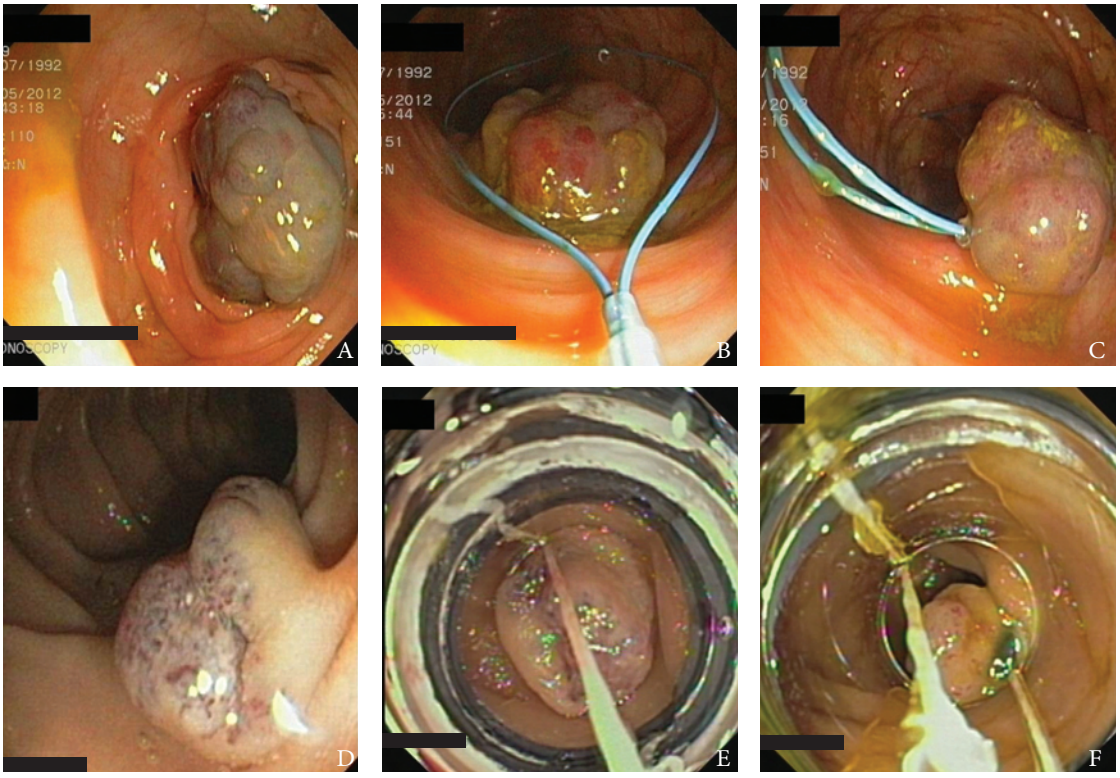

Figure 1 (A) Polypoid Blue Rubber lesion of splenic flexure. (B, C) Endoloop insertion in large bowel lesion. (D) Polypoid Blue Rubber lesion of small bowel. (E,F) Band ligation of small bowel lesions

(Fig. 1D). All lesions were ligated in one session using the band ligation technique with a multi-band ligator device (Fig. 1E,F). In order to avoid entrapment of all intestinal layers, high suction pressure was not used. In addition, two trigger cords, tied together, were used in order to match the length of the colonoscope's channel. A colonoscopy was performed and detachable snares - endoloops were inserted at the colonic lesions (Fig. 1B,C).

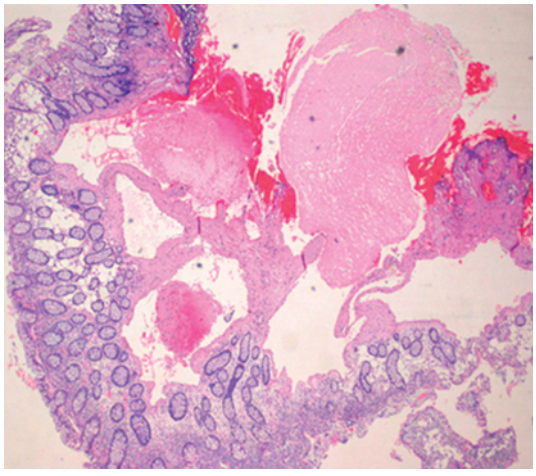

Figure 2 Histopathological section (H-E stain x 200)

A tissue specimen was obtained using the snare after endoloop insertion from one of the lesions. No bleeding occurred during the procedure. Histopathological examination revealed submucosal large blood-filled spaces lined by a single layer of endothelial cells with presence of some thrombi in the cavernous spaces (Fig. 2).

The patient made an uneventful recovery and was discharged 3 days later without any abdominal pain or signs of bleeding. Three weeks following the interventions further endoscopic evaluations were performed; these revealed scars at all sites treated with either technique. No bleeding or new venous malformations were observed (Fig. 3).

Discussion

BRBNS is a rare condition of unknown etiology and pathophysiological mechanism. Therefore, physicians are unlikely to encounter a vast number of such cases in their career, if any. Due to its infrequency and variable presentation, various treatments have been described [6-10]. Endoscopy has gained popularity in the treatment due to certain advantages over other treatment modalities. To date, no clinical studies comparing various endoscopic resection techniques have been reported, and no evidence based guidelines exist on how these lesions are best resected. The large number of

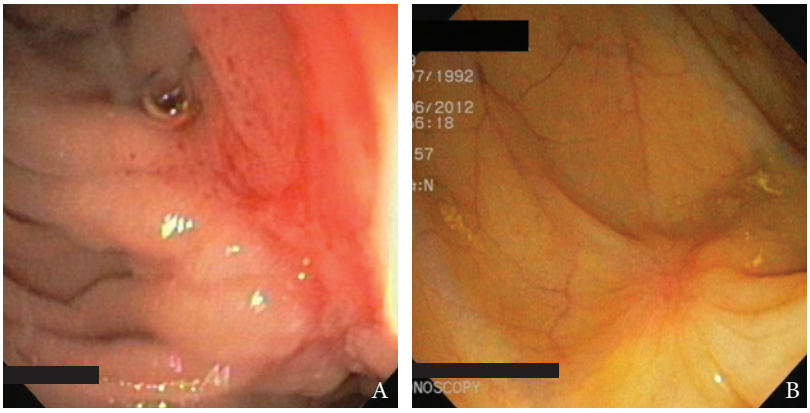

Figure 3 Posttreatment scars in (A) small bowel and (B) large bowel

endoscopic treatment methods described is likely to be an aftermath of the individual's experience, instrument availability and perceptions.

In our case, we used band ligation at the jejunal lesions for two reasons: a. the size of the lesions and b. the location. The thin jejunal wall may be at risk of thermal injury and subsequent perforation with the use of either of the techniques (snare polypectomy with electrocautery, sclerosis, laser photocoagulation, YAG laser) previously described. In addition, the use of Argon plasma electrocoagulation may lead to thermal injury of inadequate depth, reported to be 2-3 mm. In order to further eliminate the potential risk of insult into the deeper layers and perforation of the small intestine, we refrained from using high suction pressure and hence potentially avoided entrapment of intestinal wall layer deeper in the submucosa.

Endoloops were used for the polypoid and larger lesions of the large bowel (>1 cm). These would not have fitted into the cup of the band ligation device, risking incomplete resection. The endoloop strangulates the base of the polyp which subsequently leads to ischemia, necrosis-ulceration and re-epithelization, leaving behind macroscopically normal appearing mucosa or scar tissue. The endoloop was not considered suitable for the smaller jejunal lesions due to the associated risk of transaction and bleeding.

In conclusion, we report a dual technique resection method on a patient with BRBNS. The choice of resection technique depended on the lesion characteristics. Both band ligation and endoloop can safely be used for polypoid BRBNS lesions at the jejunum and colon respectively.

References

1. Gascoyen GG. Case of naevus involving the parotid gland and causing death from suffocation: naevi of the viscera. *Trans Pathol Soc London* 1860;**11**:267.
2. Massoumi H, Patel S. Blue rubber bleb nevus syndrome. *Gastrointest Endosc* 2007;**65**:1076; discussion 1077.
3. Dwivedi M, Misra SP. Blue rubber bleb nevous syndrome causing upper GI hemorrhage: a novel management approach and review. *Gastrointest Endosc* 2002;**55**:943-946.
4. Gallione CJ, Pasyk KA, Boon LM, et al. A gene for familial venous malformations maps to chromosome 9p in a second large kindred. *J Med Genet* 1995;**32**:197-199.
5. Fishman SJ, Smithers CJ, Folkman J, et al. Blue rubber bleb nevous syndrome: surgical eradication of gastrointestinal bleeding. *Ann Surg* 2005;**241**:523-528.
6. Dieckmann K, Maurage C, Faure N, et al. Combined laser-steroid therapy in blue rubber bleb nevus syndrome: case report and review of the literature. *Eur J Pediatr Surg* 1994;**4**:372-374.
7. Yuksekkaya H, Ozbek O, Keser M, Toy H. Blue rubber bleb nevus syndrome: successful treatment with sirolimus. *Pediatrics* 2012;**129**:e1080-e1084.
8. Morris L, Lynch PM, Gleason WA, et al. Blue rubber bleb nevus syndrome: laser photocoagulation of colonic hemangiomas in a child with microcytic anemia. *Pediatr Dermatol* 1992;**9**:91-94.
9. Ng EK, Cheung FK, Chiu PW. Blue rubber bleb nevus syndrome: treatment of multiple gastrointestinal hemangiomas with argon plasma coagulator. *Dig Endosc* 2009;**21**:40-42.
10. Emami MH, Haghdani S, Tavakkoli H, Mahzouni P. Endoscopic polypectomy resection of blue rubber bleb nevous lesion. *Indian J Gastroenterol* 2008;**27**:165-166.
11. Singh RA, Haber GB. Techniques in managment of nonvariceal, nonangiodysplastic gastrointestinal lesions with argon plasma coagulation, band ligation, and endoscopic clips/loops. *Gastrointest Endosc* 1999;**3**:135-139.