Case Report

Endoscopic polypectomy resection of blue rubber bleb nevus lesions in small bowel

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Blue rubber bleb nevus syndrome (BRBNS) is a rare disorder characterized by cutaneous and gastrointestinal (GI) venous malformations. The treatment of BRBNS is primarily supportive and ablative. Ablative therapy involves endoscopic or surgical treatment of GI venous malformations. We describe a 20-year-old woman who had multiple venous malformations all over the GI tract as well as cutaneous lesions. She had suffered from several episodes of melena, chronic anemia and fatigue for 10 years, which were treated temporarily by iron supplementation and blood transfusion. The endoscopic examination of the GI tract and total colonoscopy revealed multiple bluish sessile and polypoid venous malformations 2–3 cm in size throughout the GI tract. Argon plasma coagulation (APC) and polypectomy was done for all gastric and colonic lesions, respectively. Ileoscopy showed a large wide base vascular polypoid lesion at about 70 cm from the ileocecal valve with active bleeding; this was removed by snare polypectomy. One week later, she was discharged in good condition. At about 6 months’ follow up she did not report any bleeding attack. Endoscopic polypectomy can be useful in patients with large and polypoid lesions of BRBNs which are not controlled with supportive therapy. Further experience is needed to evaluate the risks versus benefits of this approach.

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In 1860, Gascoyen first described an association between cavernous hemangiomas of the skin and similar lesions in the gastrointestinal (GI) tract. In 1958, Bean further described these lesions and coined the term blue rubber bleb nevus syndrome (BRBNS).1,2 We report a case of multiple large polypoid blue rubber nevi all over the GI tract with several episodes of GI hemorrhage and severe iron deficiency anemia.

Case Report

A 20-year-old Iranian woman was admitted to Alzahra Hospital in September 2006. On admission she complained of epigastric pain, rectorrhagia and severe anemia. She had a 10-year history of sporadic episodes of melena and fatigue. She had been admitted several times earlier at the emergency service because of anemia, when she received iron supplementation and blood transfusions, which gave temporary relief. Her family history was negative for similar skin or GI disorders.

Her physical examination showed a bluish, well-margined and mildly tender skin lesion on the sole of the foot. She underwent upper GI endoscopy, which showed few small esophageal lesions, multiple gastric coin shape vascular lesions plus some coffee ground material indicating a recent upper GI bleeding. Argon plasma coagulation (APC) was done for all gastric lesions. BRBNS was diagnosed on the basis of the typical appearance and histological features of cutaneous and GI lesions. Blood transfusion was given to raise the hemoglobin to 10 g/dL. The patient was discharged in good condition. Two weeks later she was admitted again with melena and one episode of hematochezia. Upper GI endoscopy showed healed lesions in the stomach without evidence of re-bleeding. Total colonoscopy showed 4 wide base highly ectatic vascular polypoid lesions, about 2.5 cm x 2.5 cm each. Polypectomy was done for all. Only at one polypectomy site she experienced minor bleeding which was effectively controlled with APC. The terminal ileum was normal up to 30 cm of ileocecal valve. The patient was resuscitated by blood transfusion and discharged with stable hemoglobin.

About 4 weeks later, she was admitted for the third time with melena, anemia, severe fatigue and blurred vision. Upper GI endoscopy showed no more lesions or evidence of bleeding. Push enteroendoscopy showed no lesion up to the mid jejunum. Ileoscopy was done up to about 100 cm of ileocecal valve. A 2 cm x 2 cm wide base vascular polypoid lesion was seen about 70 cm of ileocecal valve with active bleeding. Snare polypectomy was done using an electrosurgical unit (Olympus PSD-10, mode blend level 2) and snare polypectome (Olympus SD-7P, BP Japan).
Colonoscopic devise used for this procedure was Olympus ClIV-U20, Extra II CV-165, scope CF-Q165L; Figure). No evidence of bleeding was seen at the site of polypectomy. Histology of the resected lesion showed cavernous hemangiomas formed by dilated capillaries with flat endothelial cells and connective tissue stroma compatible with BRBN lesion. She was discharged one week later in good condition. At 6 months' follow up she did not report any bleeding attacks.

Discussion
The treatment of BRBNS is primarily supportive and ablative.2 There is no evidence for durable beneficial effects from any drug treatment for bleeding in BRBNS.3 Ablative therapy involves endoscopic or surgical treatment of GI venous malformations. The goal of endoscopic therapy is to ablate vascular malformations in the GI tract, not only to arrest active bleeding, but also to reduce the risk of future bleeding.4,5

Venous malformations in BRBNS are usually sessile or pedunculated with a large base, hence their endoscopic removal is less commonly reported in literature.6 Teixeira et al reported a case with 3 BRBN lesions in colonoscopy, one of which was resected and 2 were treated with a purse-string suture.7 In another case report the bleeding colonic venous malformation in the transverse colon was removed by endoscopic polypectomy technique.8 In our patient, the venous malformations were sessile and large with episodic profound bleeding. We decided to perform endoscopic polypectomy for both colonic and small bowel lesions. To the best of our knowledge, there was no report of endoscopic ileal lesion resection. Since BRBN lesions predominantly occur in small bowel, this method can help in diagnosing and treatment of patients without using more invasive or costlier and time-consuming operations. So, this method can be useful in patients with large and polypoid lesions that cannot be controlled with supportive therapy, and it avoids the risks of surgery and extensive intestinal resections. Potential risks of this technique in the treatment of BRBN include bleeding, perforation and its consequences. Further experience is needed to assess the risks versus benefits of this approach.

References

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