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A Novel Endoscopic Treatment for Blue Rubber Bleb Nevus Syndrome

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Abstract

Introduction: Blue Rubber Bleb Nevus Syndrome (BRBNS), also known as Bean Syndrome, is a congenital disorder characterized by numerous, cutaneous and internal venous malformations, most commonly involving the skin and gastrointestinal tract (GI). (1) Patients with GI manifestations present with iron deficiency anemia due to recurrent GI hemorrhage from the oozing venous malformations. Due to the rarity of the condition, there are no defined guidelines on the management of BRBNS, further highlighting the need for case reports to dictate therapy.

Case Presentation: We present a case of an 83-year-old male who presented with a GI bleed due to an unknown etiology. Workup was initiated with an esophagogastroduodenoscopy (EGD) and a colonoscopy. The initial colonoscopy had poor prep and difficult visualization due to excessive bleeding. This led to extensive testing as the patient continued to have ongoing blood loss, which included a tagged red blood cell scan, angiography with attempted embolization, and magnetic resonance enterography. Finally, the patient was able to repeat a colonoscopy with appropriate prep, and findings consistent with BRBNS were identified. The patient was treated with six hemoclips of the actively bleeding spots, with resolution of his symptoms. The patient was followed for approximately two years, and has not had recurrence of GI bleed, with a stable hemoglobin.

Discussion: Blue Rubber Bleb Nevus Syndrome (BRBNS), is a congenital disorder characterized by numerous, cutaneous and internal venous malformations. The treatment of GI BRBNS is determined by the severity of the disease, taking frequency and volume of hemorrhage, as well as extent of intestinal involvement, into consideration. Medical management commonly involves the use of iron supplementation and blood transfusions. Severe cases may develop anywhere from the mouth to the anus.

The patient described in this case achieved hemostasis for approximately two years following placement of six hemoclips.

A PubMed/MedLINE literature review elucidated one other case of endoscopic hemoclipping for BRBNS, with the resolution of patient symptoms for 5 years. Endoscopic hemoclipping is a successful, novel, and clearly underutilized treatment option for BRBNS as currently there is no standard of treatment for Blue Rubber Bleb Nevus Syndrome.