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Case Report

A Case of Blue Rubber Bleb Nevus Syndrome with Isolated Colonic Lesions

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Abstract

A 65-year-old female presented with an episode of melena. Colonoscopy demonstrated diffuse colonic blebs without active bleeding. Esophagogastroduodenoscopy, capsule endoscopy, and computed tomography of the abdomen did not reveal any abnormalities. The patient required several blood transfusions. She was managed medically with oral iron no endoscopic treatment was required. This case illustrates an atypical presentation of upper gastrointestinal bleeding from isolated BLEBS.

Keywords: *Anemia; Blue Rubber Bleb Nevus Syndrome; Gastrointestinal Bleeding*

Introduction

Blue rubber bleb nevus syndrome is a rare disease, characterized by gastrointestinal and cutaneous vascular malformations that was originally described in 1860 and 1958 by Gascoyen [1] and Bean [2] respectively. Clinically, most patients present with symptoms related to iron-deficiency anemia. Rarely, patients can experience overt gastrointestinal bleeding resulting in significant transfusion requirements. The incidence of reported BRBNS is very low [3] with approximately 200 case reports published to date.

Case Report

A-65 year-old female with a past medical history of type 2 diabetes, an arterial hypertension managed by a calcium antagonist and Angiotensin-converting enzyme inhibitors, an atrial fibrillation disease treated by beta-blocker and warfarin.

She was admitted to the emergency department after she developed an episode of melena. She denied a previous history of skin lesions or a family history of similar symptoms.

Initial examination showed normal blood pressure with sinus tachycardia. Her hemoglobin was 4.8 mg/dl. She received multiple blood transfusions for her symptomatic anemia with a goal hemoglobin of greater than 10 mg/dL. Esophagogastroduodenoscopy was normal. Colonoscopy showed diffuse colonic blebs (Figure 1). The lesions were not bleeding, and were not biopsied; due to concern for bleeding. A computed tomography scan small bowel series and capsule endoscopy did not reveal any bleeding source.

During her hospitalization, the patient received six units of blood. She was managed medically with iron supplementation and hemoglobin monitoring. On follow up visit, patient had denied having any overt bleeding, and his hemoglobin was found to remain stable.

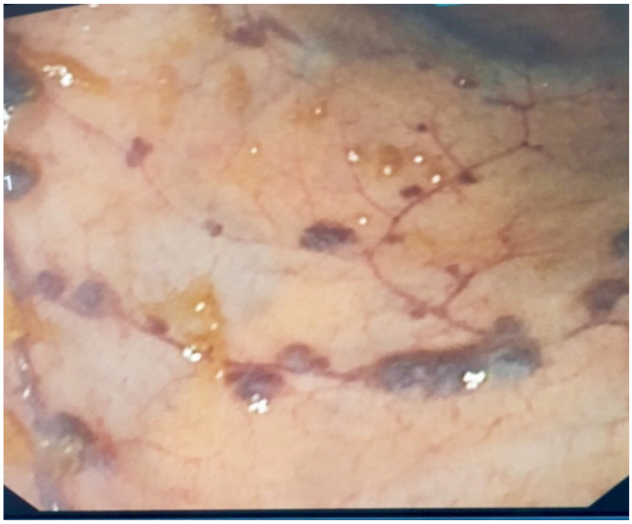

Figure 1: *Blue vascular raised lesions at transverse colon.*

Discussion and Conclusion

This case illustrates an atypical presentation of upper gastrointestinal bleeding from localized blue rubber bleb nevus syndrome, a rare vascular anomaly of unknown etiology characterized by multifocal venous malformations that affect the skin and the viscera of the gastrointestinal tract. BRNS is a sporadic disease, but familial forms have been described in the literature .In the recent studies, BRBN is caused by somatic mutations in the TEK gene, coding for TIE2 (receptor tyrosine kinase), which is involved in the PI3 K/AKT angiogenesis cascade [4].

The gastrointestinal tract is the most common organ involved in this disorder; specially the small bowel and distal colon [2]. The most frequent clinical presentation is iron deficiency anemia due to chronic bleeding, but rarely presented as melena or hematochezia [3,4].

Early diagnosis is important for the risk of life-threatening gastrointestinal bleeding. It is important to evaluate the entire gastrointestinal tract via esophagogastroduodenoscopy, colonoscopy and a video-capsule small bowel study to look for synchronous lesions. On our case, the colonic lesions was isolated.

Treatment of BRBN is symptomatic (iron and/or blood transfusions) and aims to control the digestive lesions responsible for the chronic bleeding that causes anemia [3,7]. Sclerotherapy, ligatures, laser photocoagulation have been used but require repeated general anesthesia. Corticosteroids, octreotide, interferon α, vincristine have been tried without success [5,6]. The efficacy of Sirolimus has also been reported to successfully treat skin and digestive disorders [8].

However, there is insufficient evidence regarding the duration and therapeutic benefits of these drugs proposed for the treatment of BRNS.

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