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The Curious Case of Leak Blue Blebs: A Rare Image of Blue Rubber Bleb Nevus Syndrome

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1. Case Presentation

A 78-year old man with history of hypertension, severe pulmonary hypertension, hyperlipidemia, permanent atrial fibrillation on Eliquis, CKD stage IV, iron deficiency anemia, alcohol use disorder, chronic HFrEF with EF 30%-35%, symptomatic bradycardia s/p biventricular pacemaker, coronary artery disease, STEMI status post angioplasty to mid and distal LAD in 1/2005, colon polyps, colitis of cecum, submucosal varicosities of sigmoid colon and rectum, large internal hemorrhoids, diverticulosis, and hepatic steatosis presented to the emergency department for evaluation of generalized weakness, sob and dyspnea on exertion for 6 months. His hemoglobin was notable for level of 4.7. He reported no overt signs of gastrointestinal bleeding. Pertinent examination findings included dark blue compressive papules on the hands and soles of the feet (FIG. 1).

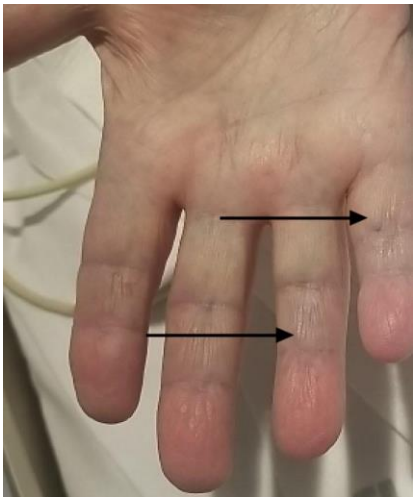

FIG. 1. **Showing blebs on hand.**

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He was transfused 3 units of packed red blood cells and was admitted for acute blood loss anemia and continued to require intermittent transfusions during his hospital stay. CT of abdomen and pelvis with contrast demonstrated no acute process. Endoscopic evaluation included an unremarkable esophagogastroduodenoscopy and colonoscopy findings of multiple, diffuse medium-sized submucosal varicosities of the sigmoid colon and rectum without bleeding (FIG. 2-4). He was treated conservatively with transfusion of packed red blood cells and iron supplementation and discharged in stable condition.

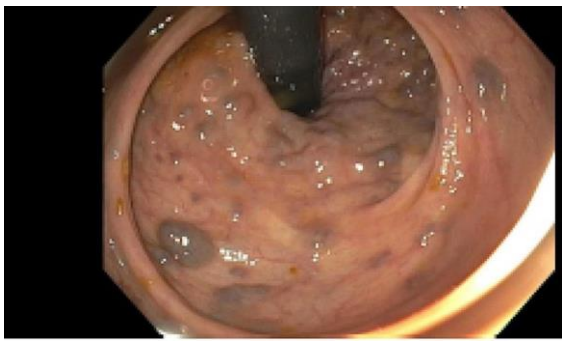

FIG. 2. **Showing submucosal of rectosigmoid junction.**

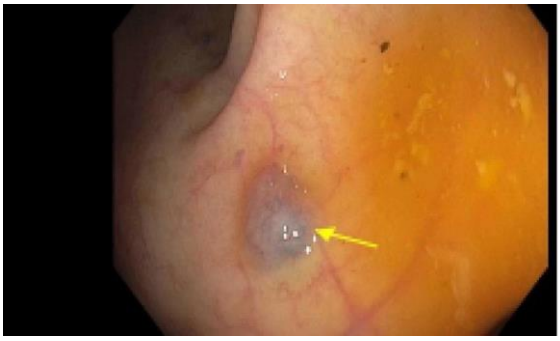

FIG. 3. **Showing submucosal varicosity of the sigmoid colon.**

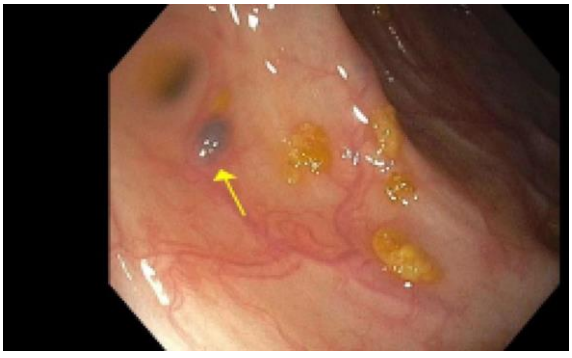

FIG. 4. **Showing submucosal varicosity of the rectum.**

2. Discussion

Given the patient's chronic anemia, skin lesions and endoscopic findings, diagnosis of blue rubber bleb nevus syndrome was made. There are no guidelines regarding the management of this condition. In the medical literature only 150 cases have been reported. Typically, patients respond well to conservative management.

Blue rubber bleb nevus syndrome (BRBN, Bean syndrome) is a rare congenital subtype of venous malformations. The dominant lesion will often develop into multiple venous malformations and will involve the skin as well as the gastrointestinal tract over time. Skin lesions are often described as multiple nevi (moles) and are often located on the palms and soles. They are characterized by multiple round lesions with bluish appearance with varying size and visual presentation; they will spring back when compressed and will refill with blood when released. Gastrointestinal sessile lesions are associated with chronic bleeding, which can progress to anemia states.

Though Blue rubber bleb nevus syndrome is a rare syndrome, the purpose of this is to serve as a reminder as it can be an overlooked cause of gastrointestinal bleeding in a person with no active signs of bleeding. A thorough workup is indicated to search for typical as well as atypical causes of blood loss in patients. Surgical intervention may be indicated depending on the degree of gastrointestinal involvement.

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