Peristomal variceal hemorrhage at the ileal conduit site due to extrahepatic portosystemic shunt

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INTRODUCTION

Peristomal variceal bleeding is a rare but known complication in a patient with portal hypertension. In patients with recurrent peristomal hemorrhage, atypical varices should be considered, and liver cirrhosis should be excluded even with normal liver function tests. Various modalities of treatment include local suturing, injection sclerotherapy, surgical shunting, and transjugular intrahepatic portosystemic shunt (TIPS) with or without embolization of varices. TIPS is the preferred modality in such patients. We report a case of a 76-year-old male who presented with recurrent ileal conduit site peristomal hemorrhage without known chronic liver disease. His liver function tests were normal, but computed tomography of the abdomen and pelvis showed liver nodularity and peristomal varices. He was diagnosed to have cirrhosis with portal hypertension and further tested positive for active hepatitis C infection. The patient’s extrahepatic portosystemic ileal conduit site shunt was successfully treated with transjugular intrahepatic portosystemic shunt and endovascular variceal coiling. This case identifies a situation where it is imperative to identify occult liver cirrhosis with portosystemic shunt as a cause of ileal conduit site recurrent stomal bleeding.

CASE REPORT

A 76-year-old male presented to the emergency department with recurrent painless bleeding from his ileal conduit site. This was his fifth visit to the hospital with similar complaints. He had a history of muscle invasive bladder cancer with peri-vesical tissue involvement, for which he underwent radical cystoprostatectomy with ileal conduit urinary diversion 7 years prior to presentation, after which he was lost to follow up. He did not take chemotherapy or radiation. He also had a history of Stage III chronic kidney disease, alcohol use of approximately 2–3 cans of beer daily for the last 40 years, and a history of medication non-compliance and non-compliance with follow-up appointments. He denied any history of nausea, vomiting, abdominal pain, back pain, blood in stools, hematemesis, or any other site of bleeding. He gave a history of fatigue, dizziness, and on and off painless bleeding from his ileal conduit site for over a year. At presentation, his heart rate was 103 beats/min, blood pressure was 126/60 mmHg, and respiratory rate was 18/min, saturating at 91% on room air. His examination was unremarkable except for gross bleed in his urostomy bag with blood clots. [His investigations revealed a hemoglobin of 3.8 g/dL (13.5–17.5),...
white blood cell count of 6.1 K/µL (4–11 K/µL), platelet count 114 K/µL (140–400), International Normalised ratio (INR) 1.38 (0.85–1.12), prothrombin time 17.6 s (12.1–15.0), total bilirubin 0.7 mg/dL (0–1.3), aspartate aminotransferase 40 U/L (14–59), alanine aminotransferase 36 U/L (11–66), and alkaline phosphatase 69 (38–126 U/L). The patient’s creatinine was elevated at presentation at 2.78 mg/dL (0.50–1.30), from his baseline creatinine of 1.32 mg/dL (estimated glomerular filtration rate = 56 ml/min/1.73 m²). After volume resuscitation, his creatinine improved to 1.4 mg/dL. He was transfused with packed red blood cells (PRBC’s). His noncontrast computed tomography (CT) of the abdomen and pelvis showed subtle nodularity of liver contour and mild splenomegaly (14.1 cm in length). Flexible cystourethroscopy through ileal conduit showed a small area of hemorrhage in the anterior portion of the conduit just deep to the abdominal wall fascia. The hemorrhagic site was fulgurated without evidence of other bleeding sites. On further examination, purple discoloration of the ileal stomal site was noticed [Figure 1]. Given his recurrent episodes of bleeding from the stomal site, purplish discoloration of the skin around the stoma, and liver nodularity, there was a high degree of suspicion of underlying liver cirrhosis with portal hypertension and extrahepatic peristomal varices. He underwent CT abdomen and pelvis with contrast, which showed heterogeneous density and enhancement of the liver, nodularity of the liver edge, and parastomal varices with mild ascites [Figure 2]. He was treated conservatively with octreotide for variceal bleeding. During the evaluation of his liver cirrhosis, his hepatitis C antibody was reactive, and his hepatitis C RNA load was 121012 IU/mL. Evaluation for hepatitis B was negative; abdominal Doppler did not show any portal venous thrombosis. His model for end-stage liver disease score at the time of TIPS procedure was 16 points, 6% estimated 3-month mortality. He successfully underwent TIPS with portosystemic pressure gradient reduction from 10 to 4 mmHg in addition to the coil embolization of ileal conduit varices by an interventional radiologist. The patient was advised to follow up with hepatology and gastroenterology for further evaluation for cirrhosis and treatment of his hepatitis C infection, respectively, and with endoscopy for evaluation of esophageal varices. He was seen in the hepatology clinic at 5 months; at which time he did not have any new episode of bleeding. He was lost to follow-up after that appointment.

**DISCUSSION**

The causes of peristomal bleeding include stomal laceration from poorly fitting appliance or rigorous cleansing or stomal varices. Peristomal varices are complications in patients with ileostomy or colostomy and underlying portal hypertension. Peristomal varices are ectopic portosystemic shunts that can develop at the mucocutaneous junction of ileostomy, colostomy, and ileal conduit site. These shunts occur between the high-pressure portal system and low-pressure systemic venous system. Peristomal variceal bleeding is rare accounting for up to 5% of all variceal bleeds. This is a well-recognized cause of peristomal bleeding in patients with underlying cirrhosis and portal hypertension. The extrahepatic variceal bleeding as a complication of stomal procedure was first described in 1968 in patients who underwent colon bypass in chronic hepatic encephalopathy. It has been increasingly recognized since patients with underlying liver disease undergo stomal procedures. The average time of development of these ectopic varices after stomal surgery varies from 19 months to 24 years as reported in one study, depending on the severity of underlying liver cirrhosis. These can also be difficult to diagnose and would require a thorough examination of the stomal site. In patients who have recurrent peristomal bleeding, it is important to exclude liver cirrhosis and portal hypertension even with normal liver function tests. Studies have demonstrated that the portal pressure gradient required for bleeding stomal varices is less than esophageal and gastric
varices. Our patient had a normal liver function test at presentation and had no evidence of esophageal or gastric variceal bleed or stigmata of decompensated liver cirrhosis at presentation, stressing the importance of excluding liver disease in these patients. Various imaging modalities such as Doppler ultrasound, CT, magnetic resonance imaging angiogram, and sometimes percutaneous angiogram have been used to aid diagnosis of ectopic varies and underlying portal hypertension. Unlike gastrointestinal variceal bleed which carries mortality risk of 30%–40%, these ectopic stomal bleeds carry mortality risk of 3%–4% with each episode. However, significant morbidity occurs due to recurrent episodic bleedings, chronic anemia requiring transfusions, and recurrent hospitalization. Treatment options for peristomal varices include conservative treatments such as local wound care, pressure dressing, suture ligation, cautery, and injection sclerotherapy with variable outcomes. These conservative therapies are associated with a high recurrence rate of up to 74% causing significant morbidity. Injection sclerotherapy has also been associated with tissue necrosis and stomal damage and is usually not recommended. Portal decompression with either surgical shunt or TIPS is often required. Surgical shunting, though effective, has 1%–50% mortality rate associated with the procedure and not always feasible in these patients. Currently, TIPS with or without embolization is a preferred treatment modality in patients with bleeding stomal varices and liver cirrhosis. TIPS decreases the portal venous pressure gradient and thereby reduces recurrent bleeding from varices.

Declaration of patient consent
The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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