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CASE REPORT | LIVER

A Bridge to Banding: Splenic Artery Embolization in Hepatosplenic Schistosomiasis

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ABSTRACT

Schistosomiasis is a parasitic infection endemic to sub-Saharan Africa. The severe form of disease, caused by deposition of *Schistosoma* eggs in the portal vein, is known as hepatosplenic schistosomiasis. We present a case of a 26-year-old woman with esophageal varices in the setting of hepatosplenic schistosomiasis. This patient underwent partial splenic artery embolization to treat thrombocytopenia secondary to splenic sequestration. After embolization and improvement of cell counts, the patient was successfully able to undergo variceal band ligation.

KEYWORDS: hepato-splenic schistosomiasis; partial splenic artery embolization; thrombocytopenia; esophageal varices; hypersplenism

INTRODUCTION

Schistosomiasis is a parasitic infection affecting approximately 236 million people worldwide. It is endemic to sub-Saharan Africa, which carries more than 90% of the global burden of disease. In this region, schistosomiasis is typically attributed to *Schistosoma mansoni* and/or *Schistosoma haematobium*.[1] Mortality is related to hepatic disease, which occurs because of deposition of *Schistosoma* eggs in the host's portal vein; this evokes an immunologic response, leading to granuloma formation and periportal fibrosis.[2,3] Resultant complications include noncirrhotic portal hypertension, leading to variceal formation and hypersplenism manifesting as pancytopenia. This severe form of disease is known as hepatosplenic schistosomiasis (HSS).[4] We present a case of a patient with thrombocytopenia secondary to HSS treated with partial splenic artery embolization (PSE) allowing for successful variceal banding.

CASE REPORT

A 26-year-old woman with a history of pancytopenia secondary to HSS presented with fatigue. The patient was born in Congo and lived in a Tanzanian refugee camp for most of her life. She immigrated to the United States in 2017, where she underwent extensive workup for splenomegaly and pancytopenia discovered during evaluation for recurrent symptomatic anemia. She was diagnosed with HSS in 2022 after a thorough workup, including esophagogastroduodenoscopy (EGD) revealing Grade III and IV esophageal varices. The patient was treated with praziquantel for schistosomiasis. Subsequent stool studies returned negative, confirming cure. She followed with gastroenterology outpatient and was started on propranolol because beta-blockers for primary prophylaxis of variceal bleeding in noncirrhotic patients with portal hypertension have proven to reduce mortality.[5]

The patient presented most recently with fatigue and was found to have worsening pancytopenia and febrile neutropenia. Laboratory test results revealed platelets 20,000/mm^3, hemoglobin 2.8 g/dL, and leukocytes 700/mm^3 from a baseline of 37,000/mm^3, 8.3 g/dL, and 1,100/mm^3, respectively. Persistent thrombocytopenia was attributed to hypersplenism from sequestration in the setting of severe portal hypertension. The patient's compliance with propranolol was questionable, given a history of nonadherence, so variceal ligation was planned; however, the thrombocytopenia needed to be corrected first. Interventional radiology was consulted and proceeded with PSE for treatment of thrombocytopenia. The postprocedural course was complicated by severe pain and splenic vein thrombosis, but given risk of bleeding in the setting of thrombocytopenia and varices, anticoagulation was held. The patient was

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eventually discharged with close follow-up. At the time of the PSE, interventional radiology also performed a liver biopsy to help guide management. Referral for liver transplant was anticipated if pathology identified liver cirrhosis, but if there was no cirrhosis, transjugular intrahepatic portosystemic shunt (TIPS) placement would be the next step. Pathology revealed normal liver parenchyma, so TIPS implantation was planned.

One month after procedure, the patient exhibited transient thrombocytosis with a platelet count of 262,000/mm^3. Leukocyte counts improved to 3,900 mm^3. After 3 months, platelet counts stabilized at 115,000/mm^3. EGD revealed Grade IV esophageal varices, for which band ligation was successfully performed, and no gastric varices were noted. Given the patient's young age and concern for noncompliance because of language barrier, lack of insurance, and history of missed clinic appointments, TIPS was ultimately not pursued. In addition, limited data exist to support improvement of pancytopenia with TIPS. The patient continues to follow with gastroenterology, with plans for repeat EGD for variceal surveillance and continued monitoring of platelet counts, given the transient nature of improved thrombocytopenia with PSE.[6]

DISCUSSION

Upper gastrointestinal hemorrhage is the primary cause of death in patients with HSS. Therefore, the most important aim in clinical management is reducing the risk of variceal bleeding. Similar to patients with cirrhosis, management of variceal bleeding can become complicated in the setting of thrombocytopenia because of limited treatment options. A study in Brazil demonstrated that thrombopoietin and reticulated platelet levels were normal in patients with schistosomiasis, emphasizing that thrombocytopenia in HSS can be attributed to retained cells in the spleen.[7] To solve this, patients may be offered splenectomy. However, morbidity ranges from 9.6% to 26.6%, and a major complication is sepsis.[8] In light of the high risk of surgery and our patient's presentation with febrile neutropenia, splenectomy was deferred.

Splenic artery embolization is another treatment for thrombocytopenia associated with hypersplenism. This procedure is known to decrease the rates of ascites and esophageal variceal bleeding while increasing hematologic indices.[9] To the best of our knowledge, PSE used to manage hypersplenism in noncirrhotic HSS has not been documented in literature. One case by Martins et al describes a patient with HSS complicated by persistent thrombocytopenia. Although PSE was planned, it could not be completed because of severe celiac trunk stenosis. Instead, the patient successfully underwent splenic radiofrequency ablation.[8] In our patient, PSE was pursued to manage thrombocytopenia and allow for eventual variceal banding. Based on a study by Zhu et al, splenic infarction rate in PSE should be limited to 50%–70% to ensure long-term efficacy in alleviating hypersplenism. Our patient had 50% infarction of the splenic artery, which resulted in appropriate response of cell counts at follow-up. The procedure does come with complications, including postoperative pain, postembolization syndrome, splenic abscesses, and splenic/portal vein thrombosis. The latter may occur secondary to decreased portal vein flow and rapid increase in platelet count after embolization.[10,11] Our patient experienced postoperative pain and splenic vein thrombosis, which have been managed on an outpatient basis.

For future steps, additional studies are needed to assess safe outcomes. A large meta-analysis conducted by Tamarozzi et al explored literature pertaining to management of HSS, including the utility of TIPS and liver transplant. TIPS has only rarely been reported in this setting, and studies regarding long-term outcomes are lacking. Therefore, the researchers could not derive conclusions from existing data. As for liver transplantation, only 1 cohort study from Saudi Arabia was identified. In the trial, 14 patients with HSS and splenomegaly received liver grafts. There was a survival rate of 75% at 1 and 5 years after transplant, comparable with that of patients undergoing transplant for other liver diseases. Three patients died within the first year after transplant, and 2 deaths were related to complications of the intervention. After transplant, neither splenomegaly nor thrombocytopenia improved.[12,13]

Our case demonstrates that partial splenic artery embolization can be used to manage thrombocytopenia in HSS. This procedure, although complicated by pain and splenic vein thrombosis, allowed for endoscopic banding of large esophageal varices by resolving thrombocytopenia. Complications can be adequately managed outpatient, making this a safe and reliable option for patients.

DISCLOSURES

Author contributions: All authors contributed to acquisition of data for the report, helped draft and revise report, reviewed and approved final version of report, and are agreeable to be accountable for all aspects of the work. N. Hassan is the article guarantor.

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Informed consent was obtained for this case report.

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