Case Report

Association of Three H - Hookworm, Hemosuccus Pancreaticus, and Hypertension (Portal) in a Patient with Melena

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Abstract
Hookworm infestations, endemic in India, are a common cause of iron deficiency anemia. Hemosuccus pancreaticus, a rare clinical condition, is due to passage of blood into the pancreatic duct possibly through a route between an aneurysm of an artery close to the pancreas and/or pancreatic duct, leading to gastrointestinal (GI) bleeding. Portal hypertensive upper GI bleed is also known since long. We report a case of a 38-year-old male with a history of alcoholism who was being investigated for GI bleeding who had concomitant hookworm infestation, hemosuccus pancreaticus as well as portal hypertension. To the best of our knowledge, this is the first report of common occurrence of hemosuccus pancreaticus and portal hypertension with hookworm infection. This case signifies the importance of infectious causes of GI bleeding to be considered even in cases where anatomic malformations or pathophysiological alterations are predominant.

Keywords: Hemosuccus pancreaticus, hookworm, portal hypertension

Introduction
Hemosuccus pancreaticus is defined as upper gastrointestinal (GI) bleeding from the ampulla of Vater through the pancreatic duct. It is most commonly associated with pancreatic inflammation, erosion of the pancreas by aneurysm, or pseudoaneurysm of the splenic artery. It occurs after the opening of pseudoaneurysm or aneurysm of splenic or gastroduodenal artery into the pancreatic duct, thus allowing blood to enter into the duct and empty from the papilla into the duodenum. Other causes of GI bleeding such as portal hypertension and infection with hookworm are well known.

We report an unusual association of portal hypertension, hemosuccus pancreaticus, and concomitant hookworm infection in a chronic alcoholic as a case of upper GI bleed.

Case Report
A 38-year-old male complaining of recurrent episodes of melena for the past 2 weeks was admitted to the emergency department of our hospital. Two days before the hospital visit, the patient had complained of the presence of blood in vomiting and had a history of recurrent complaints of upper abdominal pain often radiating to the back. His previous medical history revealed that he had suffered from chronic pancreatitis for 3 years before present admission, possibly due to alcohol abuse. He was resuscitated with the blood and intravenous fluid for melena during present admission. Ultrasonography (USG) whole abdomen revealed a moderately enlarged liver (18.5 cm × 12.3 cm) with heterogeneous echotexture, with a prominent portal vein, of size 1.3 cm; hepatic and splenic hilar collaterals were suggestive of portal hypertension. The presence of mild ascites was documented. Transabdominal USG showed echogenic pancreas with multiple, small, hyperechoic, nonshadowing foci in the pancreas suggestive of fibrotic changes. There was evidence of irregular dilated main pancreatic duct with pancreatic and intraductal calculi. These features were consistent with chronic pancreatitis. The findings were suggestive of alcoholic liver disease with portal hypertension and chronic
obstructive pancreatitis. After resuscitation, upper GI endoscopy was performed on two different occasions that showed bleeding from the second part of the duodenum with an abnormal tortuous vessel at the ampulla of Vater. This was diagnosed as hemosuccus pancreaticus, which might have caused the bleeding [Figure 1]. Serendipitously during the endoscopy, a worm was also found penetrating the mucosa of the duodenum. The worm was retrieved and referred to the laboratory for identification. It was a single intact live worm (size 12.3 mm × 0.3 mm) reddish pink, with head bent in the same direction of the body curvature with a spine at the posterior end [Figure 2a]. The buccal capsule had six teeth (four hook-like on ventral surface and two knob-like on dorsal surface) [Figure 2b]. The live worm was seen to lay eggs while examining under the microscope [Figure 2c]. Three consecutive stool samples of the patients were examined. The stool sample was dark-colored, semi-solid in consistency without any parasitic elements. Wet mount examination of stool revealed nonbile-stained ova (60–65 µm in length, 40–45 µm in width) with transparent hyaline shell membrane containing blastomeres. The above morphological features of the worm and micrometry of the ova were consistent with Ancylostoma duodenale. For further confirmation and to exclude the probability of mixed infection with Strongyloides stercoralis, the modified Harada-Mori nematode larval culture method[5] was performed, and larva of hookworm was retrieved after 1 week of incubation at ambient room temperature. However, duodenal biopsy could not be done to rule out this possibility.

The patient was a chronic alcoholic and had deranged liver functions. Other laboratory test revealed low hemoglobin (5.6 g/dL), elevated total leukocyte count (12,300/µl) with increased eosinophils (neutrophils - 54%, lymphocytes - 24%, eosinophils - 16%, and monocytes - 6%), hyperbilirubinemia (total serum bilirubin 12.5 mg% and conjugated bilirubin 11.7 mg%), alanine transaminase/aspartate transaminase (87.7/90 IU), serum alkaline phosphatase (827 IU/L), hypoalbuminemia (2.3 g%), and raised serum globulin (3.9 g%) with reversal of albumin:globulin ratio. Blood and urine cultures were bacteriologically sterile. Peripheral blood smear examination showed microcystic, hypochromic anemia, with the absence of any hemoparastises. The patient was planned for contrast-enhanced computed tomography (CECT) abdomen, but the course of illness was rapidly fatal, and he succumbed to the illness within 48 h of admission, hindering further investigative course. The cause of death was unexplained shock secondary to upper GI bleeding precipitated by chronic, alcoholic liver disease with chronic pancreatitis and portal hypertension.

**Discussion**

Upper GI bleed can occur due to varied causative factors such as anatomical, physiological, pathological, infectious, and chemical. Hemosuccus pancreaticus, a rare entity, was first coined by Sand Bloom in 1970 on detection of bleeding pancreatic duct in three cases.[1] Subsequently, more than 100 cases have been reported in English literature. Splenic artery aneurysm following pancreatitis (pseudoaneurysm) and bleeding are the primary causes of hemosuccus pancreaticus.[3,5] Rarely, pseudoaneurysm of other peripancreatic vessels such as hepatic artery, celiac artery, and gastroduodenal artery erodes into the pancreatic duct causing hemosuccus pancreaticus.[5,4] The condition is primarily seen in elderly male, usually associated with pancreatitis. This rare condition in our patient was accompanied by portal hypertension yet another common cause of GI bleeding. Portal hypertension which is often related to hepatic decompensation by underlying cirrhosis and active alcoholism is a known cause of bleeding in alcoholics and can get aggravated by other comorbid conditions.[6] Ectopic varices included in the differential diagnosis can be diagnosed with intravenous CECT or endoscopic ultrasound.[7] There was no evidence of esophageal varices in this patient; however, there were cirrhotic changes in the liver as per previous investigations. The main causes of death in such patients are due to hepatic failure, bleeding, and infections.[8]

In the present case, the exact cause of bleeding could not be zeroed due to the concomitant presence of A. duodenale.
A recent study to determine helminthic infections harbored by primary school-aged children from selected schools of Delhi, India, estimated the prevalence of hookworm to be 3.7%.\(^9\) In another study from eastern state of the country (Bihar), investigating soil-transmitted helminths in schoolchildren showed the prevalence of hookworm as high as 42%.\(^9\) The prevalence is diverse depending on the socioeconomic status, hygienic practices, open air defecation, lack of sanitation, and deworming status.\(^{10}\) Hookworm infection is acquired by penetration of the third-stage larvae (L3) through the skin. The third-stage larvae migrate through the body enter the lungs and later swallowed to reach the intestine, where it grows to adult form.\(^3\) The prevalence and intensity is higher among adult males. The inflammation in the gut triggered by feeding hookworms produces symptoms such as nausea, abdominal pain and intermittent diarrhea, progressive anemia in prolonged disease, thready pulse, cold skin, pallor, fatigue and weakness, dyspnea and in fatal cases, dysentery, hemorrhages, and edema. Adult worms though rarely seen either on endoscopy, surgery, or autopsy allow definitive identification of the species.\(^3\) Benzimidazoles, specifically albendazole and mebendazole, are the recommended antihelminthic agent against hookworm, and these agents kill adult worms by binding to the nematode’s β-tubulin and subsequently inhibiting microtubule polymerization within the parasite.\(^{11}\) Evident GI bleeding is occasionally reported in hookworm infection, and predominantly, there is slowly progressive bleeding that permits compensatory mechanisms to delay symptoms in patients. Infection with heavy hookworm burden was reported by Barakat et al. in five cases where the bleeding responded rapidly to antihelminthic treatment.\(^{12}\) Similarly, Kuo et al. have reported the successful eradication of hookworm and termination of active bleeding following mebendazole therapy.\(^{13}\) However, in our case, there were underlying causes of GI bleeding in addition to hookworm infection, leading to mortality of the patient.

In this case, all three concomitant causes might have contributed to the clinical deterioration of the patient. This case signifies the importance of infectious etiologies of GI bleeding to be considered even in cases where anatomic malformations or pathophysiological alterations are predominant.

To the best of our knowledge, this is the first report of common occurrence of hemosuccus pancreaticus and portal hypertension with hookworm infection.

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**Conflicts of interest**

There are no conflicts of interest.

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