A Rare Case of Mesenteric Ischemia (Intestinal Angina) Secondary to Superior Mesenteric Artery Thrombosis and Inferior Mesenteric Artery Atherosclerosis in a Patient with Liver Cirrhosis Secondary to Schistosomiasis: A Case Report

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Abstract. Chronic mesenteric ischemia is a rare condition accounting for less than 1 in 1,000 hospital admissions secondary to abdominal pain. Usually, at least two of the three visceral vessels need to be affected before a patient develops symptoms. We describe a patient diagnosed with decompensated liver disease secondary to liver schistosomiasis and was admitted and managed as chronic mesenteric ischemia secondary to superior mesenteric artery (SMA) thrombosis and inferior mesenteric artery (IMA) atherosclerosis. With the patient's consent, this case is presented to contribute to current knowledge about mesenteric ischemia.

Case description. A 47-year-old male diagnosed with liver cirrhosis secondary to liver schistosomiasis came in due to 1-month abdominal pain associated with 2 days' melena, diarrhea, vomiting, and 1-day disorientation. Three days post admission, he was noted with resolution of melena, diarrhea, vomiting, and disorientation; however, persistence of abdominal pain prompted consideration of chronic mesenteric ischemia and was confirmed via plain computed tomography scan. Exploratory laparotomy revealed diffuse discoloration of the small bowel but with peristalsis, thrombosis at SMA, and atherosclerosis at IMA. Heparin drip was given as management for portal vein thrombosis. He recuperated well and eventually was discharged improved.

Conclusion. Chronic mesenteric ischemia is a rare clinical symptom that occurs in less than 1 of 1,000 hospitalizations that are caused by abdominal pain. It has vague abdominal symptoms and is associated with a high mortality rate of 60% to 100%. Prognosis depends on early detection and intervention, and surgical treatment remains the option of choice.

Keywords. *Chronic mesenteric ischemia, Mesenteric thrombosis, Liver cirrhosis, Mesenteric ischemia, Intestinal angina, Liver schistosomiasis, Case report*

Introduction

Intestinal angina, or most commonly known as chronic mesenteric ischemia (CMI), is a rare condition that is gradual in onset due to the abundance of mesenteric collaterals and is oftentimes diagnosed late in its course. The frequency is very low, less than once in every 1,000 hospitalizations for abdominal pain. Patients are usually 50 to 70 years old. Females are strongly predominant, and there are other associated symptoms of atherosclerosis.[1]

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Usually at least two of the three visceral vessels need to be affected before patients develop symptoms. The diagnosis requires a high index of suspicion, and an imaging study can confirm the presence of occlusion involving the mesenteric vessels in patients, and treatment is necessary to avoid progression to infarction.[2]

The incidence of atherosclerosis in the celiac and mesenteric arteries was assessed in a series of Finnish autopsies of 120 patients. It revealed that the development of mesenteric artery stenosis is strongly associated with aging. Sixty-seven percent of subjects over the age of 80 suffered from mesenteric artery stenosis, compared with 6% of subjects under the age of 40. In the study, only one patient had bowel necrosis

during autopsy, despite occasional widespread stenotic abnormalities in the mesenteric artery.[3]

To date, there are only two reports on CMI made locally. The first one was reported in a 60-year-old female and the other one in a 74-year-old male who was diabetic, hypertensive, and dyslipidemic. We believe this case is unique since our patient is only 47 years old and with liver cirrhosis.

Case Presentation

This is a case of a 47-year-old male diagnosed with liver cirrhosis secondary to schistosomiasis for 3 years, who came in due to 1-month abdominal pain associated with 2 days' melena, diarrhea, vomiting, and 1-day disorientation.

The patient is non-hypertensive, non-diabetic, and not asthmatic. He also denies any history and exposure to pulmonary tuberculosis. Family history is unremarkable for heredofamilial diseases. He is a rice farmer, a non-smoker and an occasional alcohol beverage drinker, consuming three glasses of rhum and local beverage such as coconut wine per session approximately two times a month. He denies any illicit drug use.

One month prior to admission, he noted an onset of dull abdominal pain, but he had no fever, diarrhea, or vomiting. He self-medicated with over-the-counter hyoscine N-butylbromide tablets, which afforded temporary relief. After 2 weeks of self-medication, there was still persistence of vague abdominal pain, prompting consult and admission at a local hospital, where he was managed as a case of peptic ulcer disease and liver cirrhosis secondary to schistosomiasis. He was given unrecalled medication, which provided relief of symptoms, and hence he was discharged. Seven days after, he noted recurrence of abdominal pain associated with vomiting and worsening diarrhea, approximately ten episodes with estimated one-fourth glass volume per episode and associated with melena and disorientation, prompting admission. He was managed as a case of acute gastroenteritis with moderate dehydration, upper gastrointestinal bleeding (inactive) probably secondary to bleeding esophageal varices, hepatic encephalopathy stage 1 secondary to decompensated liver disease secondary to liver schistosomiasis.

Three days post admission, the patient's disorientation and melena had resolved but still with intermittent diarrhea and persistent abdominal pain despite intravenous antibiotics of third-generation cephalosporin (ceftriaxone). Fecalysis and stool gram stain with culture and sensitivity was done, yielding unremarkable results. The patient had stable vital signs. The abdomen was consistently flat, with hyperactive bowel sounds, soft, non-rigid, but with direct tenderness in all quadrants upon deep palpation. Suspicion of mesenteric ischemia was entertained, and abdominal X-rays taken in upright and supine views were obtained, disclosing "thumb-printing" sign of the large bowels (Figure 1). This indicates bowel edema caused by either an infectious or inflammatory process; hence, patient underwent immediate computed tomography (CT) scan of the whole abdomen with contrast.

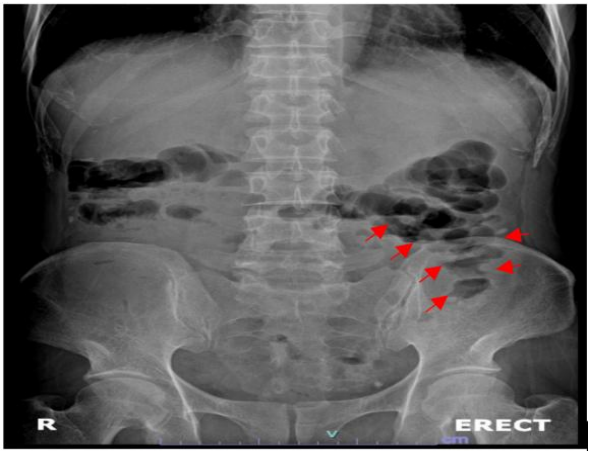

Figure 1: Abdominal upright/supine radiographic image of the patient noted with "thumb print" sign (red arrow heads)

Upon CT scan of the abdomen, thrombosis was observed in the superior mesenteric and inferior mesenteric arteries with edema and ischemia in the small intestines. There was also non-specific omental and mesenteric fat edema, diffuse liver parenchymal disease, with portal vein showing enhancing hypodense focus within the lumen, extending to the splenoportal confluence (Figure 2). High index of suspicion of CMI was further entertained.

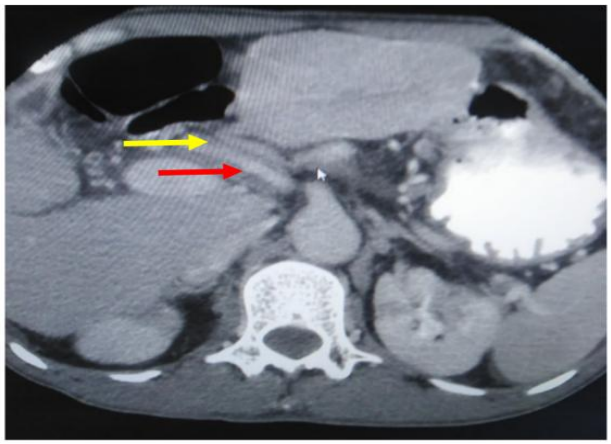

Figure 2: Abdominal CT scan with contrast revealing thrombose (red arrow) lodged in superior mesenteric artery (yellow arrow).

Immediate referral to the General Surgery Department was done, and the patient underwent immediate exploratory laparotomy. Intraoperative findings revealed ascites of 600 mL, diffuse discoloration of the small bowel 160 cm from the ligament of Treitz up to the ileocecal valve, diffuse thrombus formation on the mesenteric

vessels of small bowel (Figure 3), and a cystic lesion, probably hematoma, from the small bowel mesentery 90 cm from the ligament of Treitz. Peristalsis was still noted in the affected small bowel.

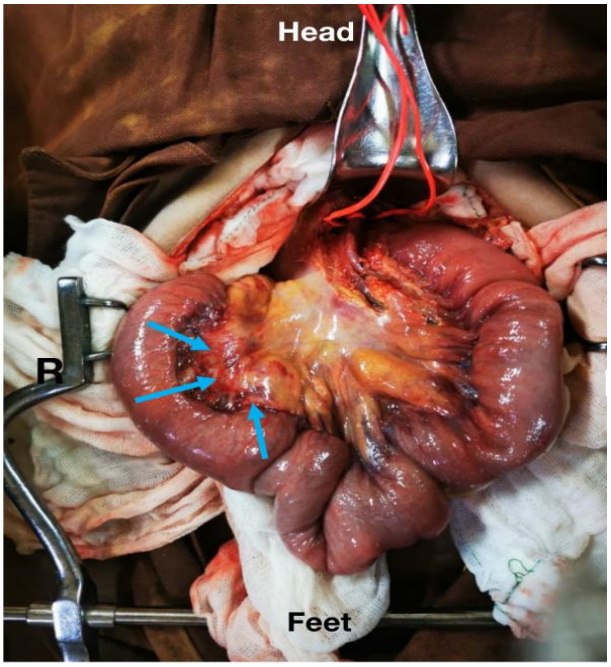

Figure 3: Diffuse thrombus formation on the mesenteric vessels of small bowel (blue arrows)

Thrombus at the origin of the superior mesenteric artery was taken through balloon-inflated thrombolectomy. Only dense atheromatous plaques were noted in the inferior mesenteric artery. The patient was then wheeled out to the post-anesthetic care unit, eventually transferred to surgical intensive care unit for close monitoring, and 2 days after was subsequently transferred to regular ward. No post-operative complications were evident. Histopathological studies were consistent with thrombus formation (Figure 4).

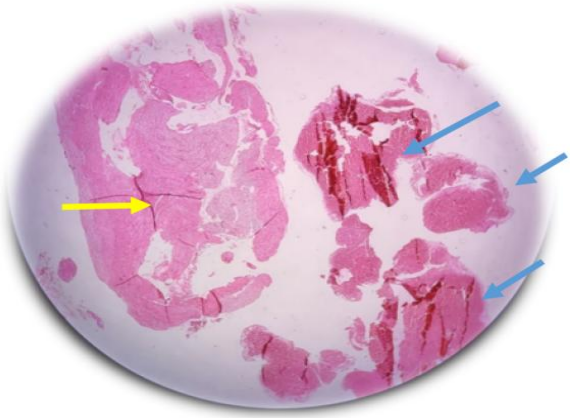

Figure 4: Biopsy result showing mesenteric vein (yellow arrow) with organizing thrombus (blue arrows) on scanner view

The concomitant portal vein thrombosis was managed with heparin drip, which was later on shifted to novel oral anticoagulant (apixaban). He was also maintained on beta-blockers (propranolol), phospholipids, and branched-chain amino acids. He recuperated well and was eventually discharged after 32 days of hospital stay, with a follow-up schedule 7 days post discharge.

Discussion

Schistosomiasis remains a general medical issue in endemic regions in the Philippines, with roughly 12 million individuals living in 28 endemic territories situated across 12 diverse topographical regions in danger for *Schistosoma japonicum* disease.[4] As the helminth lays eggs, the eggs are washed out by the blood circulation, eventually making their way to the hepatic system, where they lodge and remain viable in the liver for 3 weeks. Fundamentally, the eggs cause a moderate Th1 reaction to egg antigens. In any case, this typically advances to predominant Th2 resistant reaction to egg-inferred antigens, with later recruitment of eosinophils, granuloma development, and fibrogenesis of the liver, which advances to irreversible fibrosis.[5] Likewise, our patient resides in an area where schistosomiasis is endemic.

As cirrhosis develops, blood flow towards the liver is met with an increase in resistance; hence, there is concomitant increase in the portal pressure. In response, collateral vessels develop through the opening of prior vessels or angiogenesis. An adjustment of portal pressure is believed to be recognized first by the intestinal micro-circular vascular bed, trailed by supply routes of the splanchnic circulation. Subsequently, these vascular beds create different angiogenic factors promoting further collateral vessels development[6-11]; hence, beta-blockers were given to our patient, to decrease portal hypertension.

The presence of cirrhosis represents an expanded danger of both apoplexy and seepage in people with persistent liver illness. This is a consequence of an active disequilibrium among pro-coagulant and anti-coagulant states. Other instruments that add to this hemostatic irregularity are diminished platelet creation and expanded platelet obliteration from hypersplenism. With cirrhosis, there is diminished production of vitamin K–reliant and independent clotting elements and anticoagulants, platelet formation anomalies, and hypersplenism with platelet depletion (Figure 5).[12-16]

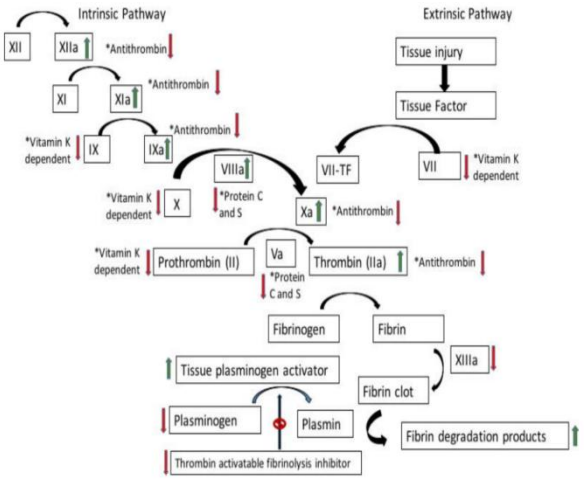

Figure 5: Coagulation cascade and the associated pathophysiologic changes that occur with cirrhosis.

The green arrow reflects the products that are increased, and the red arrow reflects the products that are decreased[12]

There is a delicate balance between bleeding and thrombose formation in normal individuals. As shown in Figure 5, in patients with liver cirrhosis similar to our case, there is decreased production of pro-coagulants, especially the vitamin K–dependent factors. However, there is also concomitant decrease in the production of thrombin inhibitors, especially the anti-thrombin. Anti-thrombin is the major thrombin inhibitor and also inhibits the factors IXa, Xa, XIa, XIIa, and VIIa+tissue factor. Hence anti-thrombin plays an immense role in the susceptibility of patients with cirrhosis to develop thrombosis.[12,17-19]

Another major factor influencing the delicate balance of hemostasis, bleeding, and thrombose formation is the presence of Virchow's triad, namely endothelial injury, stasis or turbulent flow, and hypercoagulability of the blood. In liver cirrhosis, there is increased intrahepatic resistance and hence portal hypertension. This causes disturbance to the laminar flow, causing stasis and turbulence. Stasis and turbulence therefore promote endothelial activation (enhancing procoagulant activity and leukocyte adhesion), bring platelets into contact with the endothelium, and prevent washout and dilution of activated clotting factors by fresh flowing blood and the inflow of clotting factor inhibitors.[2,17,20] With the interplay of multiple factors, patients with liver cirrhosis are not only at risk for bleeding but can also suffer from increased risks of thrombosis and/or embolism.

A study was done among 358 patients in a Dutch mesenteric ischemia expert center. One hundred thirty-eight patients were diagnosed with CMI, with a mean incidence of 9.5 (CI 95%) per 100,000 inhabitants. Majority (81%) of these patients had atherosclerotic stenosis of two or three vessels, while more than 50% had stenosis of only one vessel. Main causes of CMI were noted, and the most common was presence of atherosclerosis (mean incidence, 7.2). Other causes were median arcuate ligament syndrome (mean, 1.3) and chronic non-occlusive mesenteric ischemia (mean, 0.6). Furthermore, its occurrence is more common in the presence of more than 70% of monovascular atherosclerosis in the superior mesenteric artery (40.6%) than in celiac disease (5.6%), in contrast to our patient, who developed CMI due to thrombosis of the superior mesenteric artery.[21] Extensive literature review on local data regarding CMI only yielded two reports at the time of this study.

The magnanimous mesenteric blood supply and moderate movement of atherosclerosis permits these collateral vessels to be created. On account of these collateral pathways inside the mesenteric vasculature, patients may not experience indications of ongoing ischemia until a few significant mesenteric vessels are affected and the ongoing manifestations are brought about by the steady decline in blood stream to the digestive tracts.[2,22] Exemplary manifestations of CMI are post-prandial stomach pain associated with huge weight reduction, fear of food or of eating, queasiness, regurgitating, or loose bowel movement. The stomach discomfort or pain traditionally begins 15 to 30 minutes after food intake and regularly goes on for 30 minutes. Since complete blood supply to the digestive system can shift from 25% when fasting to 35% in the wake of eating, manifestations are more common after meals.[2,23-26] Such disease manifestations were likewise observed in our patient, who had abdominal pain, vomiting and diarrhea.

The history and physical examination, as well as a high index of suspicion, are important factors in diagnosing CMI. Although mesenteric ischemia happens rarely, the death rate is from 60% to 100%, contingent upon the source of block and prompt intervention.[23] Fortunately, up to this reporting, our patient is alive and well.

Regardless of advances in both diagnosis and treatment, rapid detection and supportive care remain critical for fruitful outcome.[23] Notwithstanding, surgical intervention remains the pillar of therapy,[26] justifying the patient's treatment modality. Surgical treatment has an unrivaled long-term patency and requires fewer re-interventions. However, it is likewise more obtrusive with more prominent morbidity and mortality contrasted with endovascular treatment. Endovascular strategies might be ideal in patients with critical co-morbidities, corresponding aortic infection, or vague symptoms.[25]

Patient's Perspective

"When I was informed of my diagnosis and the needed procedure, I was scared and hesitant. However, my doctors explained to me well the procedure, risks, and possible complications, and with great faith and fervent prayers, I finally consented and was happy that I did. Now I have recuperated, regained my full energy, and able to get back to tend our farm."

Conclusion

CMI is a rare clinical condition oftentimes presenting with vague abdominal symptoms. In addition, this condition occurred in a young patient with liver cirrhosis, further highlighting the uniqueness of this case. It has a high mortality rate of 60% to 100%, and successful outcome or prognosis depends on prompt detection and intervention. A high clinical index of suspicion and a good physical examination coupled with imaging remain a critical armamentarium. Surgical management still is the treatment of choice, although various investigations for alternative techniques are ongoing and may redirect the path on how we manage this illness in the future.

Disclosure

The authors did not receive any funding and have no conflicts of interest in conducting this study.

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