

Case Report

Strongyloides hyperinfection presenting as protein-losing enteropathy

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Abstract

Strongyloides stercoralis, a soil-transmitted helminth, affects many people in tropical and subtropical countries. It is known to cause asymptomatic infection in immunocompetent and hyperinfection and disseminated *Strongyloides* infection in immunocompromised due to autoinfection. Here, we report a case of *Strongyloides* hyperinfection presenting with protein-losing enteropathy. The diagnosis was made only with duodenal biopsy as the repeated stool examinations were negative. He was treated with ivermectin and his condition improved.

Key words

Duodenal biopsy, endoscopy, hyperinfection, protein-losing enteropathy, *Strongyloides*

Introduction

Strongyloides stercoralis is an intestinal nematode prevalent in the tropics and subtropics. It is a common cause of abdominal pain and diarrhea. Distinctive characteristics of this parasite are its ability to persist and replicate within a host for decades while producing minimal or no symptoms in individuals. Diagnosing strongyloidiasis early is important as almost all deaths are due to hyperinfection. We here report a case of *Strongyloides* hyperinfection presenting as chronic diarrhea with protein-losing enteropathy diagnosed with esophagogastroduodenoscopy biopsy of the duodenum. He showed a very good response to treatment with ivermectin. Early diagnosis and treatment prevented further worsening of the hyperinfection syndrome and saved his life.

Case Report

A 26-year-old male presented with chronic large-volume diarrhea for duration of 6 months. He did not have history

of any blood in stools. He had history of loss of appetite and loss of weight with easy fatigability. He started developing pedal edema over the last 10 days before the hospital admission. On examination, he was pale and dehydrated with angular stomatitis, glossitis, and pitting pedal edema. There was no organomegaly or palpable lymph nodes. Laboratory evaluation showed hemoglobin of 8.2 g/dl, leukocyte count of 9000 cells/cu mm with eosinophils of 7%, erythrocyte sedimentation rate of 58 mm/h, and absolute eosinophil count of 600 cells/cu mm. His liver function tests showed a total serum protein of 4.9 g/dl and albumin of 1.8 g/dl. Stool examinations repeated thrice were negative for parasites. Other biochemical investigations were normal. ELISA test for human immunodeficiency virus (HIV) and anti-tissue transglutaminase levels were negative. Peripheral smear showed microcytic hypochromic anemia. Chest X-ray showed no pulmonary infiltrates. Computed tomography of the abdomen revealed multiple mesenteric lymph nodes with prominent bowel loops. Upper gastrointestinal (GI) endoscopy revealed effacement of the mucosal folds with multiple erosions in the second and third part of the duodenum [Figure 1] from which deeper biopsies were taken. Colonoscopy showed normal mucosal and vascular pattern up to terminal ileum.

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Histopathology report from the duodenum showed ulcerated duodenal mucosa with increased cellularity in lamina propria with many of the glands containing cross section of a parasite consistent with *Strongyloides* [Figure 2]. A human serum albumin scintigraphy was done to confirm protein-losing enteropathy which showed a prominent exudation of proteins from the distal jejunum and ileum [Figure 3]. He was treated with a stat dose of 12mg of ivermectin which was repeated after 2 weeks along with supportive care with total parental nutrition and intravenous human albumin. The patient had a slow but drastic improvement in his health at the time of discharge from the hospital. His appetite improved and pedal edema came down and diarrhea had completely stopped.

Discussion

S. stercoralis is an intestinal nematode infecting about 100 million people in tropical and subtropical areas.^[1] We will discuss in short, about life cycle of *S. stercoralis* and approach to protein-losing enteropathy in this review. Infections are acquired when larvae penetrate the skin and migrate to the small intestine. The life cycle of *Strongyloides* is consisted of two parts: a free-living cycle outside the host and a parasitic life cycle with infective filariform larvae. During the free-living cycle in the soil, *Strongyloides* transform from uninfected rhabditiform larvae into infective filariform larvae, which penetrate the human skin and proceed into the submucosa, then into the venous circulation, and then toward the right heart and lungs resulting in eosinophilic pneumonitis. From the alveoli, the larvae continue to migrate up the pulmonary tree and trachea. The cough reflex pushes the larvae out of the bronchial tree and trachea. However, once the larvae reach the larynx, they are swallowed and travel to the stomach and small bowel. Inside the GI tract, *Strongyloides* larvae mature into diminutive adult females.^[2,3] Adult female worms embed themselves in the mucosa of the small bowel and produce eggs through parthenogenesis. Within the intestinal lumen, the eggs hatch into noninfective rhabditiform larvae, which are excreted, along with stool, into the environment (i.e., soil). A unique feature of nematodes, including *Strongyloides*, is their ability to cause autoinfection that is the parasite never reaches the soil; instead, it reenters the host enteral circulation (endo-autoinfection) or perianal skin (exo-autoinfection) or through the feco-oral route. Thus, parasites can remain in the human body for the remainder of the host's life.

Hyperinfection syndrome represents an acceleration of the normal life cycle of *S. stercoralis*, leading to excessive worm burden within the traditional reproductive route (the skin, gut, and lungs).^[4] Detection of an increased number of larvae in stool, sputum, and/or tissue is a hallmark of hyperinfection.^[5] Disseminated infection is the migration of larvae to organs beyond the range of the autoinfective cycle (lungs and GI tract) and is often complicated by Gram-negative sepsis.^[6] Although hyperinfection syndrome can occur in any host,

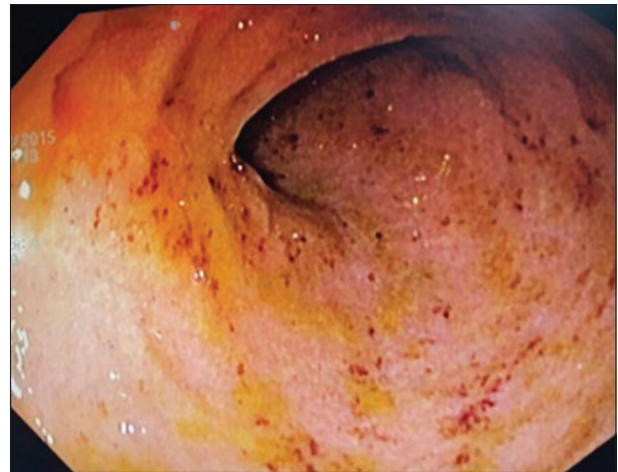


Figure 1: Upper gastrointestinal endoscopy picture showing effacement of folds in the second part of the duodenum with multiple mucosal erosions

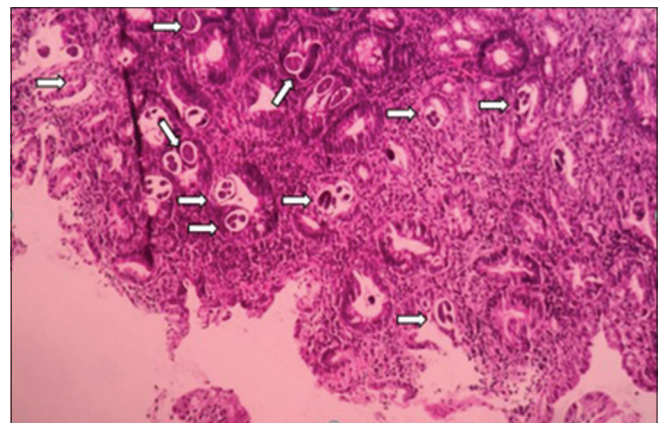


Figure 2: Histopathological section showing fragments of duodenal mucosa with a moderate increase in lamina propria cellularity by lymphocytes, plasma cells, numerous eosinophils, and neutrophils (many sections contain the parasite *Strongyloides*) (arrows pointing toward parasites) (HPE, ×10)

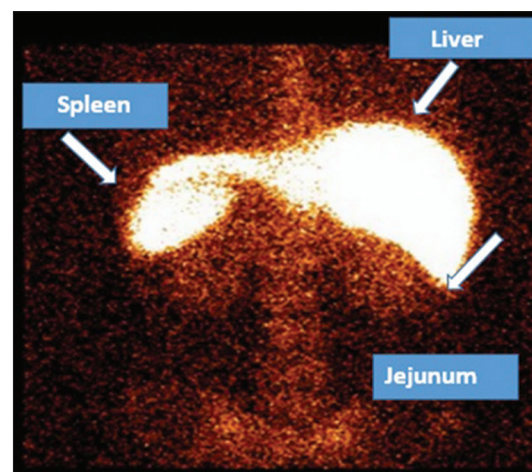


Figure 3: Human serum albumin scintigraphy demonstrating the exudation of proteins from the distal jejunum and ileum – suggestive of protein-losing enteropathy

disseminated disease occurs mainly in immune-compromised individuals.^[6] Disseminated infection is more common among the immunocompromised people such as solid organ transplants, human T-lymphotropic virus type 1 infection, HIV and in patients receiving drugs such as cyclosporine, corticosteroids, and anticancer agents.^[1,5] Disseminated *Strongyloides* infection in immunosuppressed individuals has high mortality rate from 60% to 85%.^[7] Diagnosis of *Strongyloides* hyperinfection syndrome and/or disseminated disease can be very difficult to establish. Although most studies focus on finding the parasite through stool examination, it has a yield that does not exceed 46%, even after three stool examinations.^[7]

The usual presentation of *Strongyloides* infections is GI symptoms such as abdominal pain, diarrhea, vomiting, weight loss, GI hemorrhage, fever, adynamic ileus, small bowel obstruction, and pneumonia. Very rarely, it presents as protein-losing enteropathy. The primary causes of protein-losing enteropathy can be divided into erosive GI disorders, nonerosive GI disorders, and disorders involving increased central venous pressure due to cardiac causes or mesenteric lymphatic obstruction. The initial step in the evaluation of the patient with hypoproteinemia is to exclude other more common etiologies such as malnutrition, liver, and renal diseases. After excluding, common causes for hypoproteinemia, alpha-1 antitrypsin clearance, and/or human serum albumin scintigraphy may be done to confirm the diagnosis of protein-losing enteropathy. After confirming the cause for hypoproteinemia is GI tract, the etiology for this syndrome should be evaluated. In this case, we had done gastroscopy and found erosions and ulcerations in proximal jejunum. After taking biopsy of the lesion, we were able to demonstrate the parasite in histology specimen.

Enteritis by *S. stercoralis* has been studied since the early 1960's before the endoscopic era. De Paola *et al.* classified the histopathological changes in fatal cases into three forms: catarrhal enteritis, edematous enteritis, and ulcerative enteritis.^[8] Catarrhal enteritis is a minor form characterized by mild mucosal congestion with larvae restricted to the mucous membrane. Edematous enteritis is a moderately serious form characterized by edematous thickening of the wall, swelling folds, and villous atrophy with larvae occupying lymph vessel spaces. Ulcerative enteritis is a serious form characterized by ulcers and fibrosis with larvae encountered in the entire wall. Suarez and Sánchez confirmed that plasma cell infiltration, villous atrophy, and severe duodenitis were the characteristics of severe strongyloidiasis.^[9]

Endoscopic evaluation has been recognized as an important tool for diagnosing strongyloidiasis as *S. stercoralis* colonizes in the duodenum where the larvae mature. In the endoscopic era, there have been several case reports describing endoscopic

findings of the duodenum in strongyloidiasis which can be normal mucosa, edema, erythema, erosion, swollen folds, fine granule, tiny ulcer, polyps, hemorrhage, megaduodenum, deformity, and stenosis.

The first-line therapy for strongyloidiasis is ivermectin which achieves eradication rates of approximately 80%. Other effective agents include thiabendazole and albendazole. Often, a single course of treatment is insufficient. Thus, if symptoms do not resolve after the initial therapy, it is imperative that diagnostic studies be repeated to determine the persistence of the *Strongyloides* infection and whether a second course of therapy should be given.

Conclusion

Our patient was a normal immunocompetent person who presented with protein-losing enteropathy with chronic diarrhea and peripheral eosinophilia and repeated stool examinations negative for parasitic infections. Finally, he was diagnosed with *Strongyloides* hyperinfection with the help of duodenal biopsy. He was treated with two doses of ivermectin and supportive treatment as he had a picture suggestive of hyperinfection. He showed a remarkable improvement on follow-up for the last 3 months without any further complications.

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Conflicts of interest

There are no conflicts of interest.

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