Case report

A case of Strongyloides Stercoralis induced duodenitis and pancreatitis

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A B S T R A C T

Strongyloidiasis is endemic in tropical and sub-tropical regions however cases of strongyloidiasis have been reported in temperate climates. Corticosteroid use, immunosuppression, infection with human T-lymphotropic virus type 1 (HTLV1), and chronic alcohol use are the most common and well-established risk factors for strongyloidiasis. Due to Strongyloides stercoralis characteristic features of hyperinfection and dissemination, it can potentially cause a lethal infection in an immunocompromised individual. Strongyloidiasis is predominantly asymptomatic, however some unusual manifestations of strongyloidiasis include duodenal obstruction, ileus, reactive arthritis, ascites, hepatic lesions, and pancreatitis. Here we present a case of a 47-year-old-St. Lucian female who was found to have duodenitis and pancreatitis secondary to Strongyloides stercoralis in the setting of underlying HTLV-1 infection and chronic alcohol use.

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Introduction

Strongyloidiasis is an opportunistic infection caused by the helminth Strongyloides stercoralis and is most transmitted via the soil. Strongyloides stercoralis has an estimated prevalence of 30–100 million cases worldwide \cite{1,2}. The disease is endemic in tropical and sub-tropical regions however cases of strongyloidiasis have also been reported in temperate climates \cite{2}. In developed countries, S. stercoralis infection is very rare however some occupations such as mining, and farming have been associated with an increased risk of infection \cite{3}. In Europe and the United States, infection occurs in clusters and predominantly affects individuals such as tourists and travelers, and military personnel returning home from endemic areas, or in immigrant populations that have migrated from endemic areas \cite{3}. Strongyloidiasis is predominantly asymptomatic, however typical manifestations of strongyloidiasis include dermatological, pulmonary, and gastrointestinal symptoms including hemorrhage. Hyperinfection encompasses a spectrum that includes high parasitic burden, and clinical manifestations can include intestinal damage, respiratory distress, sepsis and/or meningitis due to enteric bacterial superinfection. Hyperinfection due to strongyloidiasis carries a mortality rate up to 87\% \cite{4}. Any alteration in an individual's immune status can lead to hyperinfection and dissemination of parasites that can present with severe symptoms such as ileus, duodenal obstruction, and pancreatitis. Glucocorticoid use, chronic alcohol use and HTLV-1 infection are two of the most common etiologies that are associated with hyperinfection \cite{4,5}. There is also a clear established relationship between chronic alcohol use as a risk factor for acquiring strongyloidiasis.

Case presentation

A 47-year-old female from the Caribbean Islands presented to our facility with complaints of worsening epigastric pain for one day. Patient had intermittent epigastric pain for many years that was diagnosed as peptic ulcer disease. Current episode of epigastric pain was 10/10 on the pain scale, radiating to the right shoulder, unrelieved by acetaminophen, and associated with nausea and multiple episodes of non-bloody, non-bilious, watery emesis. She did not have weight loss but did report subjective fevers, and chills. Her last alcoholic beverage use was reported to be two days prior to presentation. Her medical history was significant for systemic hypertension and gastroesophageal reflux disease (GERD) and an umbilical hernia repair surgery. The patient was born in St. Lucia, in the Caribbean Islands and moved to the United States 5 years ago. She did not report any recent travel outside the state of New Jersey, or outside the US. She denied any sick contacts and reported no recent antimicrobial use. She did however report drinking alcohol (mostly beer) every day for many years and last use (3–5 shots of Italian liqueur) three days prior to presentation.

On physical examination, the patient appeared in mild distress secondary to epigastric pain and showed some signs of dehydration.

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Her vital signs demonstrated high blood pressure of 147/90 mmHg. The abdominal examination was significant for tenderness in the epigastric region, with no peritoneal signs and a visible well healed umbilical hernia scar. Laboratory workup revealed leukocytosis with white blood cell of 11,000/μL (n = 4500–11,000/μL) including neutrophil 73% (n = 36–66%) and eosinophil 4% (n = 0–8%), a mild normocytic anemia with hemoglobin 11.1 g/dL (n = 12–15 g/dL). Other laboratory review was significant for an alanine aminotransaminase (ALT) 148 Unit/L (n = 13–56 Unit/L), aspartate aminotransaminase (AST) 314 Unit/L (n = 15–37 Unit/L), an alkaline phosphatase 134 Unit/L (n = 45–117 Unit/L), a normal total bilirubin 0.5 mg/dL (n = 0.2–1.0 mg/dL) and a lipase level 3230 Unit/L (n = 73–393 Unit/L). Additional workup showed hemoglobin (Hb) A1c 5.8% and a procalcitonin level 43.3 ng/mL.

Initial imaging on workup included abdominal ultrasound which revealed a dilated common bile duct (CBD) at the level of the pancreatic head with no gallstones or gallbladder wall thickening. An abdominal computed tomography (CT) with contrast showed dilated CBD but reported no mass or intrahepatic biliary dilatation. (Fig. 1). Subsequently, an upper endoscopy was performed that showed reflux esophagitis and gastritis. Tissue sample from the duodenum was consistent with duodenitis. Microscopic analysis of the submucosal layers was consistent with Strongyloides stercoralis and inflammatory cells (Fig. 2). The patient was also noted to have reactive HTLV-I/II antibodies, elevated IgG4 of 134 mg/dL (n = 4–86 mg/dL) as well as trichrome stain ova and parasites positive for Strongyloides stercoralis rhabditiform larvae. Further diagnostic and therapeutic evaluation with Endoscopic Retrograde Cholangio Pancreatography (ERCP) was performed which revealed a moderately dilated main bile duct secondary to distal stricture and thus a biliary stent was placed. Given the present nematode in stool studies, the cause of patient’s duodenitis and pancreatitis was suggested to be due to Strongyloides stercoralis and patient was diagnosed with strongyloidiasis. Once the diagnosis of strongyloidiasis was established, ivermectin, 200 μg/kg per day for 2 days, was given. Her appetite improved and the vomiting subsided. Prior to discharge, a stool ova and parasites sample were collected and was reported later as positive for Strongyloides stercoralis rhabditiform larvae. By day 3, patient started to feel much better and was subsequently discharged home. Patient returned for a follow-up visit a week later and complained of abdominal pain. An additional two-day course of ivermectin 15 mg oral twice a day was then prescribed with significant alleviation of symptoms.

**Discussion**

Strongyloidiasis is a unique helminth infection caused by *Strongyloides stercoralis* which is endemic in tropical and sub-tropical regions such as the Caribbean and is also present in low endemicity in temperate climates [5]. *S. stercoralis* is transmitted via skin contact with contaminated soil whereas less common modes include fecal-oral and person-to-person transmission.

There are several etiologies that have been associated with *S. stercoralis* infection. Steroid use, immunosuppression, infection with human T-lymphotropic virus type 1 (HTLV1), and chronic alcohol use are the most common and well-established risk factors for strongyloidiasis [5]. The disease often manifests in individuals treated with corticosteroids, although often underlying HTLV-1 infection is also present as a risk factor [6]. A case series of strongyloidiasis reported a prevalence of 27% of individuals with concomitant HTLV-1 infection thereby suggesting a strong association of *S. stercoralis* infection with HTLV-1 [7]. It is hypothesized that these etiologies (HTLV 1 infection & corticosteroid use) cause immunosuppression that allows *S. stercoralis* to systematically multiply in uncontrollable fashion, also known as hyperinfection.

Numerous cases have suggested an association of hyperinfection and dissemination of *S. stercoralis* in immunocompromised patients [1]. Mora et al. reported 11 cases of *S. stercoralis* hyperinfection in individuals with systemic lupus erythematosus (SLE) receiving steroids and other immunosuppressive agents [8]. Similarly, chronic alcohol use can also predispose an individual to *S. stercoralis* infection. In a study on chronic alcoholic population in Brazil, the estimated frequency of infection was noted to be 6 times greater than in healthy controls [9].

Many individuals infected with *S. stercoralis* remain asymptomatic, however symptoms pertaining to pulmonary and gastrointestinal involvement may be present in symptomatic individuals [5]. Acute symptoms of *S. stercoralis* infection include localized skin irritation (at the site of entry), edema or urticaria whereas chronic infection is predominantly asymptomatic however symptoms such as nausea, vomiting, diarrhea, weight loss, abdominal pain, gastrointestinal hemorrhage, cough, fever, and dyspepsia may be present [10]. Some unusual manifestations of strongyloidiasis include duodenal obstruction, ileus, reactive arthritis, ascites, hepatic lesions and pancreatitis [11]. Duodenitis due to strongyloidiasis is an uncommon finding however few cases of *S. stercoralis* induced duodenitis have been reported in literature. Randale et al. reported a
case of chronic duodenitis due to strongyloidiasis that was diagnosed via EGD proven biopsies [10]. Sheth et al. reported a case of gastric and duodenal ulcer caused by *S. stercoralis* in an immunocompetent host [12].

Additionally, a rare association of *S. stercoralis* with pancreatitis has also been reported in literature. In 2008, Perez-Jorge and Burdette reported a case of pancreatitis in an 81-year-old female in which the only etiologic factor identified was *S. stercoralis* recovered during endoscopic retrograde cholangiopancreatography [13]. Pijls and associates reported a case of pancreatitis in a 30-year-old patient who was infected with *S. stercoralis* and who then developed biliary stenosis due to an enlarged pancreatic head [14]. *S. stercoralis* predominantly persists in human duodenum and upper jejunum however they can often migrate via the biliary tract to cause cholangitis and/or pancreatitis [15].

Early diagnosis of Strongyloidiasis carries significant importance as it can reduce morbidity and mortality by reducing risk of hyperinfection, autoinfection and dissemination. Definitive diagnosis of *S. stercoralis* usually depends on presence of larva in stool however in some cases it might be false negative due to low intestinal worm load. Multiple studies have suggested to obtain 3–7 serial stool samples that significantly increase diagnostic sensitivity from 50% to 95% [1]. As *S. stercoralis* predominantly persists in human duodenum and upper jejunum endoscopic evaluation can help in establishing strongyloidiasis. EGD has been recognized to be an important tool for diagnosing strongyloidiasis [10]. Multiple case reports have identified maturing larvae colonizing in crypts and glands of duodenal mucosa [10,16].

Our patient was a chronic carrier of *S. stercoralis* which she had most likely acquired from St. Lucia in the Caribbean, which is a known endemic area with a prevalence of 2.9% [17]. Our patient's history of chronic alcohol abuse and the concomitant HTLV-1+ infection rendered an immunocompromised state that predisposed our patient to hyperinfection by *S. stercoralis* that resulted in severe duodenitis. In our patient, *S. stercoralis* first habituated in the duodenum causing duodenitis, which was confirmed via EGD biopsy. Furthermore, we postulate that the migration of the nematode into the biliary tract would have caused pancreatitis. Once the diagnosis was established our patient was immediately treated with Ivermectin with prompt improvement in symptoms by day 3.

With this case we would like to emphasize the importance of early diagnosis and prompt treatment of strongyloidiasis as the case fatality rate of hyperinfection syndrome in patients with diminished cellular immunity is between 50% and 86% [18]. Additionally, we would also like to emphasize on maintaining a low threshold for EGD and biopsy in immunocompromised patients who present with gastrointestinal symptoms as delay in establishing diagnosis of strongyloidiasis can exponentially increase morbidity and mortality.

**Conflicts of interest**

No conflict of interests.

**Acknowledgement**

None.

**Footnotes**

The authors have no conflicts of interest to declare. The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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**References**

[1] Jones N, Cocchiarella A, Faris K, Heard SO. Pancreatitis associated with Strongyloides stercoralis infection in a patient chronically treated with corticosteroids. J Intensive Care Med 2010;25(3):172-4. https://doi.org/10.1177/ 0885565410365099. PMID: 20444734

[2] Buonfrate D, Bisanzio D, Giorli G, Odermatt P, Fürst T, Greenaway C, et al. The global prevalence of Strongyloides stercoralis infection. Pathogens 2020;9(6):468. https://doi.org/10.3390/pathogens9060468. PMID: 32545787; PMCID: PMC7349567.

[3] Toledo R, Muñoz-Antolín C, Esteban JG. Strongyloidiasis with emphasis on human infections and its different clinical forms. Adv Parasitol 2015;88:165–241. https://doi.org/10.1016/bs.apar.2015.02.005. Epub 2015 Mar 23. PMID: 25911368

[4] Nutman TB. Human infection with Strongyloides stercoralis and other related Strongyloides species. Parasitology 2017;144(3):263–73. https://doi.org/10.1017/S003118201600834. Epub 2016 May 16. PMID: 27181117; PMCID: PMC5563389

[5] Makker J, Balar B, Niazi M, Daniel M. Strongyloidiasis: a case with acute pancreatitis and a literature review. World J Gastroenterol 2015;21(11):3367–75. https://doi.org/10.3748/wjg.v21.i11.3367. PMID: 25805946; PMCID: PMC4363769.

[6] Dykie A, Wijesinghe T, Rabson AB, Madugula K, Farinas C, Wilson S, et al. Human T-cell leukemia virus Type 1 and Strongyloides stercoralis: partners in pathogenesis. Pathogens 2020;9(5):11094. https://doi.org/10.3390/pathogens9110904. PMID: 33137906; PMCID: PMC7692131.

[7] Kockza Charles Philip, Hindy Pierre, Goodman Adam, Gress Frank. Strongyloidiasis. Eur J Gastroenterol Hepatol 2012;24(7):860–2. https://doi.org/10.1097/EJG.0b013e3283543ae0

[8] Moras C, Segami ML, Hidalgo JA. Strongyloides stercoralis hyperinfection in systemic lupus erythematosus and the antiphospholipid syndrome. Semin Arthritis Rheum 2006;36(3):135–43. https://doi.org/10.1016/j.semarthrit.2006. 06.001. Epub 2006 Sep 1. PMID: 16949135.

[9] de Oliveira LC, Ribeiro CT, Mendes Dde M, Oliveira TC, Costa-Cruz JM. Frequency of Strongyloides stercoralis infection in alcoholics. Mem Inst Oswaldo Cruz 2002;97(1):119–21. https://doi.org/10.1590/S0074-02762002000100021.

[10] Randale A, Dani A, Chawhan S, Meshram S, Tathe S, Kumbhalkar D. A case report of Strongyloides stercoralis duodenitis in an immunocompromised patient. Parasitol United J 2015;8:127–9.

[11] Koprowski A, Nutman TB, Strongyloides: a neglected tropical disease. Infect Dis Clin N Am 2019;33(1):135–51. https://doi.org/10.1016/j.idc.2018.10.006. PMID: 30712758; PMCID: PMC6337075.

[12] Sheth S, Assof F, Hallit R, Sison R, Afzali M, Spira R, et al. Strongyloides: the cause of multiple gastrointestinal ulcers in an immunocompetent individual. Case Rep Med 2014;2014;346256. https://doi.org/10.5184/crmed.2014.346256. [Epub 2014 Feb 4. PMID: 24668845; PMCID: PMC3932278].

[13] Perez-Jorge EV, Burdette 3D. Association between acute pancreatitis and Strongyloides stercoralis. South Med J 2008;101(7):771–2.

[14] Pijls NH, Yap SH, Rosenbusch G, Prener H. Pancreatic mass due to Strongyloides stercoralis infection: an unusual manifestation. Pancreas 1986;1(1):90–3.

[15] Tanaka T, Hirata T, Parrott G, Higashiarawa M, Kinjo T, Kinjo T, et al. Relationship among Strongyloides stercoralis infection, human T-cell lymphotropic virus type 1 infection, and cancer: a 24-year cohort inpatient study in Okinawa, Japan. Am J Trop Med Hyg 2018;94(2):365–70. https://doi.org/10.4269/ ajtmh.15-0556.

[16] Kakati B, Sang S, Heif M, Caradine K, McKnight W, Aduli F. Strongyloides duodenitis: case report and review of literature. J Natl Med Assoc 2011;103(1):60–3. https://doi.org/10.4103/0027-9684/15(3)30426-7.
[17] Kurup Rajini, Hunjan Gurup. Intestinal parasites in St Lucia: a retrospective, laboratory-based study. J Rural Trop Public Health 1924:9.

[18] Jegadeesan R, Sundararajan T, Jain R, Karnik T, Ardasenov Z, Sidorenko E. Strongyloides duodenitis in an immunosuppressed patient with Lupus Nephritis. Kans J Med 2018;11(1):1–9. [Published 2018 Feb 28].