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Case report

CASE REPORT

Hepatobiliary strongyloidiasis presenting as an ampullary lesion on esophagogastroduodenoscopy/endoscopic ultrasound

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Abstract

Strongyloidiasis is an intestinal infection caused by the parasitic nematodes of the *Strongyloides* species, most commonly *Strongyloides stercoralis*. We report a case of a 66-year-old immigrant male from Haiti who presented with complaints of diarrhea and an unintentional 80-lb weight loss over the past 5 years. Stool examination was positive for strongyloidiasis. Following albendazole therapy, esophagogastroduodenoscopy (EGD) showed a unique ampullary lesion. Histopathology of the ampullary lesion showed reactive epithelium with *Strongyloides* larva. In addition, endoscopic ultrasound (EUS) detected a large pancreatic cyst. Both these findings were absent on EGD 5 years previously, prior to the onset of his symptoms. This paper documents a rare case of an ampullary lesion and pancreatic cyst secondary to hepatobiliary strongyloidiasis in a non-Human Immunodeficiency Virus (HIV) patient. We review the epidemiology, life cycle, clinical presentation and treatment of strongyloidiasis.

Key words: strongyloidiasis; *Strongyloides stercoralis*; ampulla of Vater; pancreatic cyst; hepatobiliary; endoscopy

Introduction

Strongyloidiasis is a parasitic disease that affects 100 million people worldwide [1, 2]. With a unique life cycle and route of infection, strongyloidiasis can give rise to infection that may persist for decades. Individuals may be asymptomatic or experience mild cutaneous and abdominal symptoms such as urticarial, pruritus, nausea, diarrhea and abdominal pain [1]. Due to its global prevalence, strongyloidiasis should be considered as a differential in patients from endemic areas, regardless of the severity of their symptoms. Furthermore, most cases of strongyloidiasis have been documented in Human Immunodeficiency Virus (HIV) or immunocompromised patients. Herein, we present a rare case of an ampullary lesion and pancreatic cyst secondary to hepatobiliary strongyloidiasis in a non-HIV patient.

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Case

A 66-year-old Haitian male with a medical history of hypertension and benign prostatic hyperplasia presented with complaints of chronic diarrhea for several months. Diarrhea was associated with night sweats and an unintentional 80-lb weight loss over the past 5 years. He denied fever, chills, myalgias, pruritus, cough, wheezing, chest pain and shortness of breath. Our patient had emigrated from Haiti approximately 10 years previsouly; however, he reported visiting Haiti numerous times since then. Physical examination was unremarkable. Laboratory findings were significant for an elevated eosinophil count of 8%. The patient was negative for HIV. Other laboratory abnormalities included anemia (hemoglobin 11.7 g/dL, mean corpuscular volume 97 fL), basophilia (1.2%), hypoalbuminemia (3.2 g/dL) and hypoproteinemia (5.4 g/dL). Alanine aminotransferase, aspartate aminotransferase, alkaline phosphatase and total bilirubin were within normal reference ranges. He was diagnosed with strongyloidiasis based on the identification of stool larvae. During his hospital course, albendazole therapy was initiated and administered for 4 days.

Given the patient's history of weight loss (80 lbs) and night sweats in the setting of positive TB Quantiferon test, computed tomography (CT) scan of the chest/abdomen was performed. Pancreatic and common bile duct dilations were incidentally noted on abdominal CT. Chest CT scan showed the presence of pulmonary apical scarring, reticulonodular changes and prominent inguinal lymph nodes. A QuantiFeron test was positive. Taken together, these findings were diagnostic for latent tuberculosis infection. The patient denied ever having been diagnosed or treated for active or latent tuberculosis. In addition, he denied pulmonary symptoms such as cough, wheezing and dyspnea. Abdominal CT revealed a dilated pancreatic duct, intrahepatic and extra-hepatic biliary duct dilation without choledocholithiasis, and a pancreatic cyst.

The patient was referred for esophagogastroduodenoscopy (EGD) and endoscopic ultrasound (EUS) to further characterize his symptoms of diarrhea, weight loss and the abnormally dilated pancreatic duct, which was noted on CT scan. EGD revealed gastritis and a suspicious ampullary lesion (Figure 1). Multiple ampullary lesion biopsies followed by histopathology showed reactive epithelium with larva of *Strongyloidiasis* (Figure 2). TB gram stain and culture was not performed on the ampullary lesion. On EUS, the ampullary wall appeared hyperechoic. The main pancreatic duct was dilated to 3.5 mm at the body and tail of the pancreas. An anechoic intraductal pancreatic main duct cyst measuring 8.9 mm x 13.8 mm was visualized at the head of the pancreas (Figure 3). Fine needle aspiration showed normal levels of CEA, amylase and lipase, and was negative for malignant cells. Of note, the patient had an EGD and colonoscopy about 4 years previously, which was unremarkable.

A few weeks after discharge, the patient followed up after completion of albendazole therapy and reported complete resolution of diarrhea and night sweats. He also noted a 10-lb weight gain in the span of 2 weeks. The patient was continued on isoniazid and vitamin B6 therapy for latent TB. MRI abdomen/MRCP was performed 3 months later, which noted unchanged biliary and main pancreatic duct dilatations with tapering to normal caliber distally. No discrete biliary filling defect was identified. An unchanged 0.8-cm cyst in the tail of the pancreas was noted—a finding without overtly complex features. No ampullary lesion was identified.

Discussion

Strongyloidiasis is endemic to warm and humid climates, with an estimated global prevalence of 100 million people [1, 2]. According to the Centers for Disease Control and Prevention, the percentage of infected individuals in the USA can vary anywhere from 0 to 46.1% in certain immigrant populations, and between 0 and 6.1% in other populations such as travelers or military personnel [3, 4].

Strongyloidiasis is often contracted percutaneously when individuals come into contact with soil containing infectious *Strongyloides stercoralis* filariform larvae [5]. Strongyloidiais is most often associated with walking barefoot, agricultural activities, and poor sanitation and sewage disposal. Hence, resource-poor tropical and subtropical environments provide ideal conditions for infection and transmission [6, 7]. Rhabditiform larvae must develop into filariform larvae in order to be infectious. This process may occur in the soil or within the bowel of an infected individual. Once the filariform larvae penetrate the skin or mucous membranes, they enter the bloodstream and travel to the pharynx, where they can be swallowed [8]. In this way, larvae enter the gastrointestinal system, where they mature into adult worms.

Risk factors that predispose individuals to superinfection include prior infection with human T-lymphotropic virus 1 (HTLV-1) and impaired immunity [9]. In fact, both infection with HTLV-1 and impaired immunity, either secondary to glucocorticoid therapy, immunosuppressive drugs, alcoholism, malnutrition, organ transplantation, HIV/AIDS or hematologic malignancies such as leukemia or lymphoma, can lead to infection followed by hyperinfection syndrome, where large numbers of filariform larvae are produced and disseminated [9–14].

The clinical presentation of strongyloidiasis infection is nonspecific, with symptom severity depending on the level of host immunity and exposure to filariform larvae. Healthy individuals may be asymptomatic or experience mild cutaneous and abdominal symptoms. In disseminated disease, parasites may be found in organs beyond the skin, gastrointestinal tract and pulmonary system, causing meningitis, gram-negative or polymicrobial sepsis and/or pneumonia [15]. Fluctuating eosinophilia is common in mild disease, whereas severe disease often lacks this diagnostic clue [16]. Elevated IgE levels are also expected in the majority of cases [17].

Regardless of symptom severity, strongyloidiasis must be treated to avoid the possibility of hyperinfection syndrome or disseminated disease. Ivermectin, albendazole and thiabendazole are treatment options used throughout the world. Oral ivermectin, the drug of choice, has been proven to be more effective and better tolerated than albendazole [18]. Unfortunately, many endemic countries do not have access to ivermectin, which is also used in cases of disseminated disease. Eradication of infection is usually confirmed by repeat stool examination 2–4 weeks after drug therapy.

Isolated cases report that strongyloidiasis is associated with nodular lesions in biliary ducts and bronchi, the former resulting in obstructive jaundice and the latter resulting in asthma-like symptoms secondary to an obstructive lung pattern [19, 20]. Another case report described biliary obstruction resulting from papillary stenosis caused by *S. stercoralis* [21]. While most of these cases report strongyloidiasis in immune-compromised patients, our paper describes a rare case of strongyloidiasis in an immune-competent patient.

Despite the potentially problematic location of the parasitic mass on the ampulla, our patient did not present with

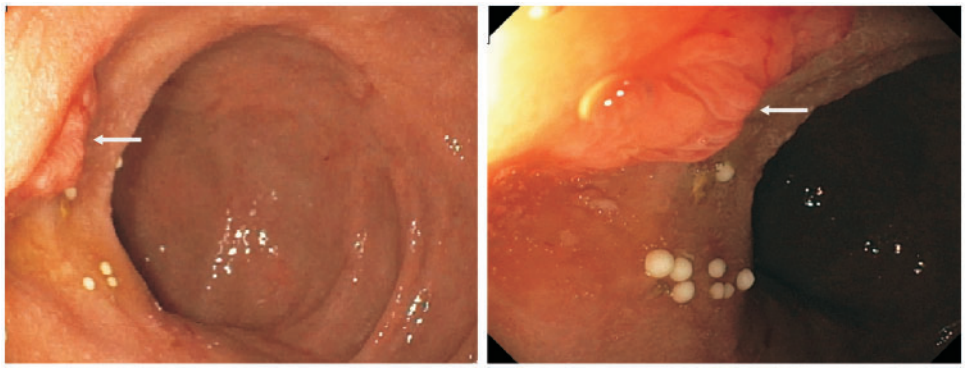

Figure 1. Upper endoscopy reveals ampullary lesion.

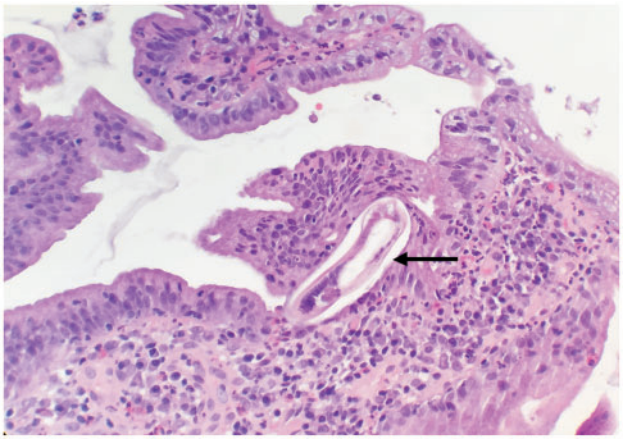

Figure 2. Reactive epithelium with larva of Strongyloidiasis.

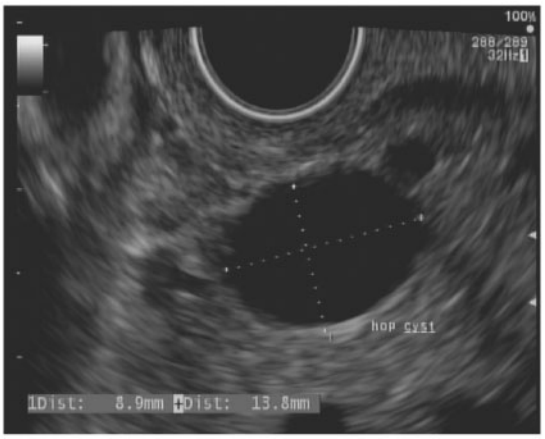

Figure 3. Endoscopic ultrasound view of pancreatic cyst (8.9mm x 13.8mm) in the head of the pancreas.

obstructive symptoms. Furthermore, a relationship between strongyloidiasis and certain pancreatic manifestations has been suggested. For instance, one case highlights a possible relationship in which strongyloidiasis was associated with pancreatic cystadenocarcinoma [22]. Other cases have described the occurrence of pancreatitis secondary to *Strongyloides* infection [23]. In our patient, the pancreas was harboring a large intraductal pancreatic main duct cyst. Though we cannot confirm that the patient's pancreatic cyst was related to strongyloidiasis infection, the patient's clinical picture was highly suggestive.

Conclusion

We report a very rare and peculiar finding of a non-obstructing ampullary lesion and pancreatic cyst in a non-infected HIV patient. Our patient was diagnosed with strongyloidiasis and treated with albendazole. Strongyloidiasis has the potential to become a life-threatening infection if not treated properly. Due to international travel and immigration from endemic areas, physicians must be aware of the infectious etiology of diarrhea, night sweats and weight loss. Clinicians should bear in mind that immunocompromised patients are at risk for developing strongyloidiasis.

Authors' contributions

E.O., D.R. and G.S. designed the report; E.O., A.K., D.R., M.R. and G.S. collected the patients' clinical data; P.X. acquired images; D.R. and A.K. wrote the paper; E.O., M.R. and G.S. edited the paper for intellectual content. All authors approved the final version of the manuscript.

Conflicts of interest

None declared.

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