Case Report: *Bacteroides*-Associated Pylephlebitis in a Patient with Strongyloidiasis

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Abstract. Strongyloidiasis is associated with Gram-negative bacteremia. Septic portal vein thrombosis or pylephlebitis is a rare but serious complication of intra-abdominal infection, and it is often associated with *Bacteroides* bacteremia. We present the first report of pylephlebitis with *Bacteroides* bacteremia associated with underlying *Strongyloides stercoralis* infection and briefly review the management of septic portal vein thrombosis.

INTRODUCTION

Infection with the nematode *Strongyloides stercoralis* presents with a spectrum of disease from asymptomatic infection to the hyperinfestation syndrome. Gram-negative bacteremia is associated with strongyloidiasis,¹ particularly the hyperinfestation syndrome, in which it can be a fatal complication. Bacteremia in this setting is often polymicrobial and predominantly with Gram-negative gastrointestinal organisms, such as *Escherichia coli* and *Bacteroides fragilis*.² Pylephlebitis is a rare complication of intra-abdominal sepsis, and it is diagnosed on the basis of a new finding of a portal vein thrombus in the context of fevers and bacteremia.³ *B. fragilis* is the single most commonly isolated organism from the blood in reported cases of pylephlebitis.⁴ We present the case of a patient with polymicrobial bacteremia (including *B. fragilis*) secondary to underlying strongyloidiasis with subsequent development of septic portal venous thrombosis.

PATIENT

A 61-year-old Ghanaian male presented with a 6-week history of fevers, night sweats, increased frequency of stool, central abdominal pain, and 14 kg of weight loss. He had visited relatives in Ghana for 3 months before the onset of symptoms. He was based mainly in urban Kumasi; however, he visited rural villages in the Ashanti region of Ghana. He was well on his return to the United Kingdom. On examination, he was febrile at 38°C; however, there were no localizing signs of a septic source. Blood results revealed a marked anemia, with hemoglobin of 7.6 g/dL (mean corpuscular volume (MCV) = 93 fL). C-reactive protein was 217 mg/L, and he had neutrophilia (7.7×10^9) and mild eosinophilia (0.5×10^9). Renal and liver functions were normal. He was treated empirically for intra-abdominal sepsis with intravenous ceftriaxone and metronidazole and received 2 units of blood. Blood cultures drawn on admission, before the administration of antimicrobials, grew *B. fragilis*, *Eggerthella lenta*, and *Myroides odoraminatus*. Stool culture was negative, and no parasites were seen on microscopy. The presence of mixed anaerobic bacteria in the bloodstream pointed to a gastrointestinal source of sepsis; therefore, a CT scan of the abdomen and pelvis was ordered. This revealed numerous small liver cysts thought to be benign by appearance and no other significant pathology.

Upper gastrointestinal tract endoscopy was performed, which showed duodenitis. D1 and D2 biopsies showed small

bowel mucosa with villous blunting and surface foveolar gastric metaplasia as well as chronic inflammation of the lamina propria. A colonoscopy revealed the presence of diverticulosis and a single polyp, which was subsequently shown to be histologically benign. He was, by this point, noted to have rising peripheral blood eosinophilia (1.0×10^9 , which was 10% of the total white cell count). Given his travel history, *Strongyloides* serology was requested, which was strongly positive on enzyme-linked immunosorbent assay (ELISA; optical density [OD] = 1.967, cutoff for positivity = 0.461). As a result, oral ivermectin (200 mg/kg) was added to his therapy. Human T-lymphotropic virus (HTLV) I and II and human immunodeficiency virus (HIV) antibodies were negative.

Given the ongoing clinical concern of possible gastrointestinal malignancy, he proceeded to a positron emission tomography–computed tomography (PET–CT) scan, which showed marked fluorodeoxyglucose (FDG) avidity throughout the biliary tree (Figure 1). A magnetic resonance (MR) scan of the liver was, therefore, advised, and this revealed a portal vein thrombus extending into the superior mesenteric vein (Figure 2). A thrombophilia screen, including protein C, protein S, prothrombin 20210 mutation, prothrombin time, and Factor V Leiden, was negative. He was treated with a 4-week course of antibiotic therapy (intravenous ceftriaxone and metronidazole for 2 weeks followed by 2 weeks of oral coamoxiclav) and anticoagulation with warfarin for the portal vein thrombus.

He has since made a complete clinical recovery, with resolution of anemia and fevers, and he has returned to his baseline weight. Eosinophilia also resolved. His eosinophil count was 1.0×10^9 on starting ivermectin; by day 3 of treatment, it was 0.8×10^9 , by day 7 of treatment, it was 0.7×10^9 , and by day 9 of treatment, it was 0.6×10^9 . When next tested (15 days after starting ivermectin), eosinophil count had normalized to 0.4×10^9 , and it was 0.1×10^9 when taken at a follow-up visit 1 month later.

DISCUSSION

To our knowledge, this is the first report of pylephlebitis identified in association with strongyloidiasis. *Strongyloides* hyperinfestation syndrome has long been associated with Gram-negative bacteremia,⁵ and it is likely that our patient's bacteremia was related to *Strongyloides* infection given the travel history, positive serology, and eosinophilia at the time of positive blood culture.

Additionally, *Bacteroides* bacteremia seems to be associated with pylephlebitis (i.e., septic thrombosis of the portal vein). Universally fatal before the antibiotic era,⁶ the condition still carries significant morbidity and mortality (32% recorded in one case series).⁴ It is most commonly described as occurring

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FIGURE 1. Fused CT/PET image showing markedly increased branching uptake in the liver.

secondary to diverticulitis; however, it has been reported with appendicitis and cholecystitis.⁷ It is nearly always associated with bacteremia, which is often polymicrobial, and *B. fragilis*, which was isolated in our patient, is the single most commonly isolated organism.⁴ This is postulated to be secondary to the induction of procoagulant macrophage activity by *B. fragilis*.⁸ Duration of antimicrobial therapy is not well-established, but

FIGURE 2. MR imaging of the liver (post-contrast image). The arrows highlight that vessels are low signal (black), suggesting thrombus.

4–6 weeks has been recommended.⁴ Anticoagulation is often prescribed with the goal of preventing thrombus extension. There are little data to support its use; however, in one review of 100 cases, 35 cases received anticoagulation. There was 6% mortality in those receiving anticoagulation compared with 22% mortality in those who did not receive anticoagulation.⁷ Another retrospective review of 44 patients suggested better outcomes in those who were anticoagulated.⁹

This case simultaneously highlights the need to consider strongyloidiasis as an underlying cause of intra-abdominal sepsis in a patient with relevant exposure and septic portal venous thrombus (pylephlebitis) as an important and potentially dangerous complication of intra-abdominal sepsis.

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