Case Report

Diverticulitis-induced pylephlebitis possibly misdiagnosed as biliary duct obstruction

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Case: A 59-year-old Asian man presented to our emergency department with hypogastrium pain, loss of appetite, and diarrhea. On admission, he was hypotensive and jaundiced. Laboratory test results revealed thrombocytopenia, hypercreatininemia, and hyperbilirubinemia. Color Doppler sonography showed no blood flow in the right and left branches of the portal vein, which seemed similar to biliary obstruction. Enhanced computed tomography showed portal vein thrombi, consistent with pylephlebitis; a broad-spectrum antibiotic and an anticoagulant were administered.

Outcome: The patient died of multiple organ failure 22 h post-admission. An autopsy revealed suppurative thrombi in the portal vein, multiple liver abscesses, and diverticulitis in the sigmoid colon.

Conclusion: Pylephlebitis, a rare complication of intra-abdominal infections, is associated with high rates of morbidity and mortality. Ultrasonography findings mimic those of biliary obstruction. Enhanced computed tomography is useful for diagnosing this condition.

Key words: Cholestasis, intra-abdominal infections, sepsis, thrombophlebitis, ultrasonography

INTRODUCTION

Pylephlebitis is a suppurative thrombophlebitis of the portal vein and its branches. It is a rare complication resulting from intra-abdominal infection or hypercoagulability, such as malignancies or coagulation disorders.1,2 Pylephlebitis was reported as a source of liver abscess for the first time in 1846.3 In the past, intra-abdominal infections, such as appendicitis and diverticulitis, were major common causes of pylephlebitis, but a recent study noted that biliary infection is currently the most common cause.3 Clinical features are non-specific, and the diagnosis of pylephlebitis is often difficult. Pylephlebitis remains a life-threatening illness, although antibiotic use is widespread; the mortality rate, which is as high as 30-50%, is related to late diagnosis due to its non-specific findings and rare occurrence.4

Here, we present a case of pylephlebitis complicated with sigmoid colonic diverticulitis in a patient who died of septic shock.

CASE

A 59-YEAR-OLD ASIAN MAN, who had alcoholic liver disease due to heavy alcohol consumption, presented to our emergency department with a 20-day history of hypogastrium pain, loss of appetite, and diarrhea.

The patient’s vital signs on presentation were: body temperature, 36.1°C; heart rate, 91 b.p.m.; blood pressure, 80/52 mm Hg; respiratory rate, 32 breaths/min; and SpO2, 98% on ambient air. He had jaundice, and his abdomen was tender at the hypochondriac region. Laboratory test results were as follows: white blood cell count, 4,600/lL; platelet count, 48,000/lL; prothrombin time, 40.5%; activated partial thromboplastin time, 37.4 s; fibrinogen/fibrin degradation products, 31.2 µg/mL; D dimer, 5.4 µg/mL; serum creatinine, 3.2 mg/dL; blood urea nitrogen, 205 mg/dL; total bilirubin, 9.1 mg/dL; alkaline phosphatase, 1,047 U/L; and γ-glutamyltransferase, 154 U/L. The blood culture later indicated Klebsiella pneumoniae and Streptococcus constellatus infection.

Color Doppler ultrasonography showed the absence of blood flow in the right and left branches of the portal vein, which seemed similar to biliary dilation (Fig. 1). Initially, the tentative diagnosis was acute obstructive suppurative cholangitis, according to laboratory and echographic findings. However, enhanced computed tomography (CT)
showed that the right and left branches of the portal vein were not enhanced and that there were multiple cystic lesions in the entire liver, not consistent with biliary obstruction but consistent with pylephlebitis and multiple liver abscesses (Fig. 2). Moreover, CT revealed multiple diverticula of the sigmoid colon with partial wall thickness (Fig. 2).

The patient was admitted to the intensive care unit, where a broad-spectrum antibiotic (meropenem) and an anticoagulant (heparin) were administered i.v. Because the patient became hypoxic and had anuria, ventilator management and continuous renal replacement therapy were started. Moreover, massive and continuous infusion of noradrenaline and vasopressin were administered due to septic shock. However, the patient died of multiple organ failure 22 h after admission.

An autopsy revealed suppurative thrombi in the portal vein, multiple liver abscesses, and diverticulitis in the sigmoid colon (Fig. 3). The definitive diagnosis was pylephlebitis and multiple liver abscesses due to sigmoid colonic diverticulitis.

**DISCUSSION**

In the present case, ultrasonography showed no blood flow in the portal vein, which seemed similar to biliary duct dilation, and the patient died of septic shock due to pylephlebitis and liver abscesses complicated with diverticulitis, despite intensive care.

Clinical features of pylephlebitis are non-specific. A retrospective single-center study reported that this condition occurred in patients with a median age of 57 years, and 83% of patients were men. Common symptoms are abdominal pain and fever; 50% of patients with pylephlebitis have anergic abdominal pain, and 30% of these have fever. Laboratory test results usually show leukocytosis and normal liver function, even in cases with severe sepsis or liver abscess. Blood culture tests are positive in 80% of the patients with pylephlebitis, and the common pathogens are *Bacteroides fragilis*, enterobacteria, and streptococci. *Klebsiella pneumoniae* is significantly related to liver abscess.

Risk factors for pylephlebitis include immunosuppressive conditions, such as diabetes mellitus, HIV infection,
This patient had an alcohol addiction, with no liver cirrhosis observed at autopsy. Although alcoholism has not been reported as a risk factor, alcohol use disorders are related to immunocompromised states. Therefore, alcohol addiction might have contributed to pylephlebitis in the present case.

Abdominal Doppler echography typically shows the absence of blood flow in the portal vein; however, this finding can mimic biliary duct dilation, as in the present case. Contrast-enhanced CT, which shows contrast defects of the portal vein, is not only more useful for definitive diagnosis but also for evaluating liver abscess and intra-abdominal infection. In late cases presenting with jaundice and severe sepsis, the diagnosis is often made at autopsy.

Antibiotic therapy is the most important for treating pylephlebitis; however, the use of anticoagulants is controversial. Once the diagnosis is made, a broad-spectrum antibiotic should be immediately administered for empiric therapy, until blood culture results are obtained. Antibiotic therapy should be given for 4 weeks to patients without liver abscess and for 6 weeks to those with liver abscess. If the size of the liver abscess is >3 cm, drainage should be considered. A retrospective study showed that in 70% of patients with portal and mesenteric vein thrombosis, early anticoagulation achieved complete or partial thrombus resolution; however, the efficacy of anticoagulation is not clearly defined in pylephlebitis. In addition to the treatment of pylephlebitis, treatments of the infection source, such as with surgery and drainage, are also important. In the present case, the enhanced CT showed abnormal findings in the sigmoid colon. However, the CT finding did not enable the diagnosis of diverticulitis with abscess. If it had, a treatment strategy that included surgery or drainage might have been carried out.

Pylephlebitis can be a fatal complication of intra-abdominal infection even if antibiotics are administered. The diagnosis is difficult because of its non-specific clinical features and echography mimicking biliary obstruction. Therefore, it is important to diagnose accurately this rare but life-threatening complication.

CONFLICT OF INTEREST

None.

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