

## Pylephlebitis as complication of late diagnosis of cholangitis: case report and review of the literature

### *Pilephlebitis como complicación del diagnóstico tardío de colangitis: reporte de un caso y revisión de la literatura*

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#### Keywords:

Portal vein,  
cholangitis, sepsis,  
liver abscess.

#### Palabras clave:

Vena porta,  
colangitis, sepsis,  
absceso hepático.

#### ABSTRACT

Pylephlebitis is the septic thrombosis of the portal vein or its tributary branches, which occurs because of an intraabdominal or pelvic infectious focus. The clinical manifestations can be very nonspecific, from asymptomatic to septic shock and liver failure. The knowledge of this pathology allows the diagnosis that is based on a high clinical suspicion supported by imaging tests, tomography is the study of choice. It is an infrequent complication, but with a high mortality rate. We present the case of a 37-year-old patient with pylephlebitis secondary to choledocholithiasis.

#### RESUMEN

La pilephlebitis es la trombosis séptica de la vena porta o de sus ramas tributarias que ocurre como consecuencia de un foco infeccioso intraabdominal o pélvico. Las manifestaciones clínicas pueden ser muy inespecíficas, desde asintomáticas hasta choque séptico e insuficiencia hepática. El conocimiento de esta patología permite el diagnóstico que se basa en una alta sospecha clínica, el cual se apoya en las pruebas de imagen; la tomografía es el estudio de elección. Es una complicación infrecuente, pero con una elevada tasa de mortalidad. Se presenta el caso de una paciente de 37 años de edad con pilephlebitis secundaria a coledocolitiasis.

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Received: 30/07/2018  
Accepted: 21/11/2018



### INTRODUCTION

Pylephlebitis refers to the septic thrombosis of the portal vein or its tributary branches, which results from uncontrolled intra-abdominal or pelvic infection, regions neighboring or drained by the portal system.<sup>1</sup>

It was first described in 1846 by Waller, who discovered it as originating in liver abscesses at an autopsy.<sup>2,3</sup> It begins as thrombophlebitis of small mesenteric veins that spread to the portal venous system and liver. Thrombosis of the mesenteric veins can occur, leading to mesenteric ischemia and intestinal necrosis. If the suppurative process is extended, abscesses are formed, the most frequently appearing in the right liver lobe.<sup>4</sup>

Pylephlebitis affects patients in a wide age range, from 20 days to 77 years, with an average presentation of 42 years.<sup>5</sup> 0.6% of all intra-abdominal infections are complicated by pylephlebitis.<sup>6</sup> It was classically described as a complication in cases of appendicitis; however, with antibiotic therapy, the incidence of pylephlebitis of this cause has decreased. Currently, diverticulitis and pancreatitis are the main causes,<sup>3,5</sup> although it can also be associated with inflammatory bowel disease, intestinal perforation, necrotizing pancreatitis, pelvic infections, hyper-coagulable states, trauma or abdominal surgery, cirrhosis, or contiguous infection (e.g. cholangitis).<sup>7</sup>

It is possible to find bacteremia associated with pylephlebitis, which is usually

**How to cite:** Ángeles-Gaspar D, Telich-Tarriba JE, Leyva-Sotelo L, Guevara-Valmaña OI, Rendón-Medina MA. Pylephlebitis as complication of late diagnosis of cholangitis: case report and review of the literature. Cir Gen. 2019; 41(2): 114-118.

polymicrobial, the most frequently involved microorganisms are *Streptococcus viridans* (24%), facultative Gram-negative bacilli as *Escherichia coli* (21%), and anaerobes as *Bacteroides fragilis* (12%).<sup>3</sup>

The clinical manifestations are nonspecific, they include general malaise, fever, generalized abdominal pain, and nausea. Jaundice is unlikely and involves advanced forms with significant liver damage.<sup>3</sup>

Its diagnosis requires high clinical suspicion and the support of imaging tests, ultrasound, and abdominal CT scan, which have sensitivity and specificity. Abdominal ultrasound can demonstrate echogenic material within the lumen of the vessels, or liver abscesses. Doppler ultrasound can provide information on the alteration in porto-mesenteric flow.<sup>8</sup> CT scan is the method of choice; intravascular air can be an initial finding of the disease.<sup>2</sup> CT scan also allows identifying intra-abdominal or pelvic infectious foci, as well as a distinctive hypodensity in portal-mesenteric thrombosis, compared with the hyperdensity of the contrast medium.<sup>9</sup> The anatomical locations of septic thrombosis by frequency are the right or left portal veins, the hepatic veins, the superior mesenteric or inferior, and the splenic vein.<sup>3</sup>

The most common complications are of liver abscesses, reported in up to 37% of patients.<sup>5,9</sup> It can also be complicated by

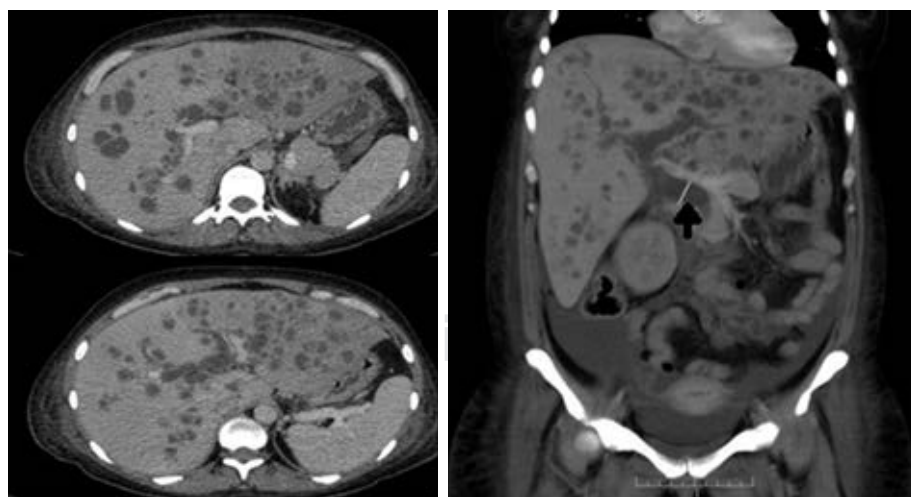
intestinal ischemia or portal hypertension.<sup>5,10</sup> Currently, even with adequate antimicrobial therapy and anticoagulation, it still has a significant mortality rate, which varies from 11 to 32%.<sup>3,5</sup>

This article aims to have pylephlebitis in mind as a possible diagnosis in a septic patient with nonspecific abdominal symptoms, which are the common denominator. It is an infrequent complication, with a high mortality rate. Diagnosis and treatment, which includes antibiotics and, in specific cases, anticoagulation or surgery, must be done early because morbidity and mortality depend on it.

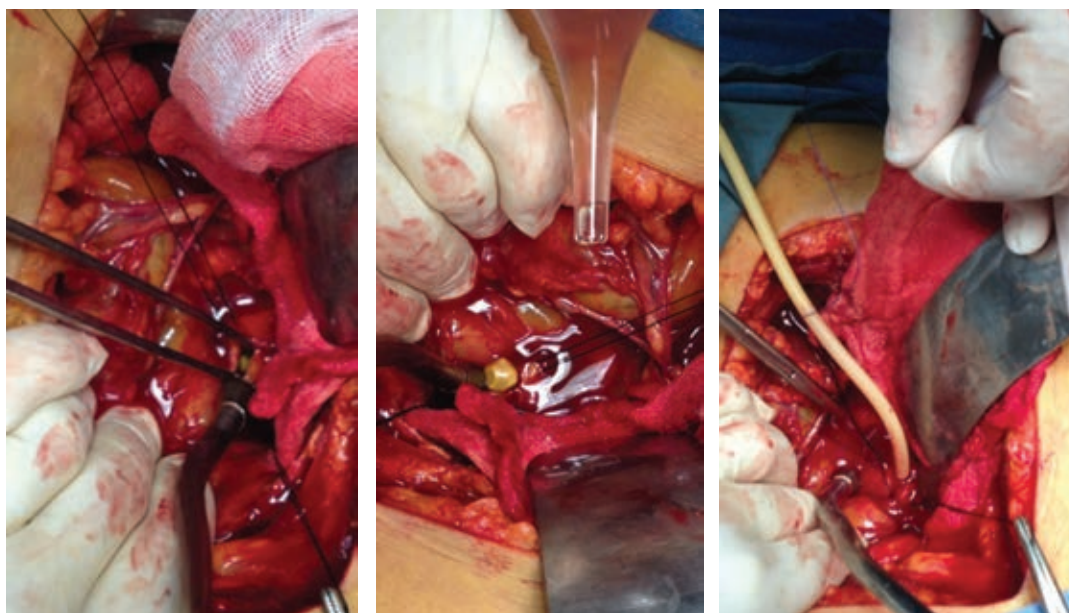
### PRESENTATION OF A CASE

A 37-year-old female decided to attend to the Emergency Department with profuse epistaxis, treated by nose packing, she referred intermittent pain in the right hypochondrium of a year's duration, which exacerbated five days before admittance, accompanied by asthenia, adynamia, and jaundice of skin and sclera. Likewise, she had had a fever in the previous 48 hours, and history of laparoscopic cholecystectomy 18 months before.

On physical examination, she was found alert, with generalized jaundice, a heart rate of 104 BPM, a temperature of 36.5 °C, and BP of 110/70 mmHg. The abdomen was painful



**Figure 1:** CT scan with coronal and cross sections, where multiple liver abscesses, intrahepatic and extrahepatic, and bile duct dilation (arrow) and thrombosis of the portal vein are shown.



**Figure 2:**

*Bile duct exploration with choledochotomy, stone extraction, and placement of a T tube.*

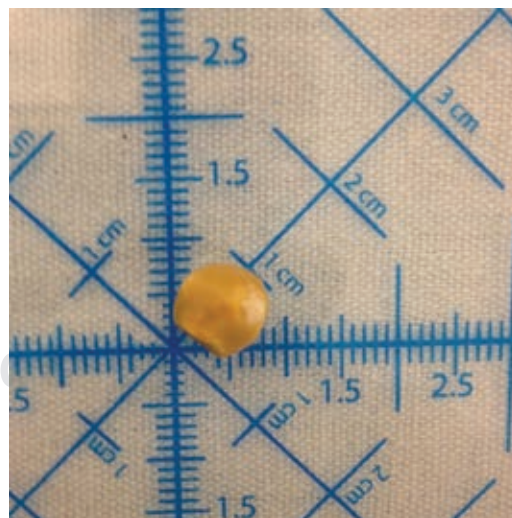
on medium and deep palpation, with greater intensity in the right hypochondrium, positive, tympanic Murphy sign, with liver dullness 2 cm below the subcostal border, and no evidence of peritoneal irritation. The rest of the examination showed no alterations.

Laboratory tests showed hemoglobin of 8.8 mg/dl, leukocytosis of 61,700/mm<sup>3</sup> mainly of polynuclear cells (95%), 23,000 platelets/ $\mu$ l. Her liver function tests showed total bilirubin of 24.0 mg/dl, direct bilirubin 19.7 mg/dl, and indirect bilirubin of 4.3 mg/dl. ALT 67 U/l, AST 90 U/l and alkaline phosphatase 861 U/l, creatinine 1.8 mg/dl, urea 77 mg/dl, PT 42.8 sec, PTT 39.8 sec, an INR of 3.64.

An ultrasound of the liver and bile ducts was done, which revealed the absence of the gallbladder, a heterogeneous liver with multiple abscesses dilation of the intrahepatic and extrahepatic bile ducts, and a common bile duct 13 mm in diameter. CT scan showed cystic liver images, intra- and extrahepatic dilation, significant bile duct dilation, and portal vein thrombosis (Figure 1). With these findings, the diagnosis of pylephlebitis associated with the formation of multiple liver abscesses, as a complication of cholangitis was made.

Because endoscopic retrograde cholangiopancreatography (ERCP) was not available, the

patient was admitted to the operating room for an open exploration of the bile duct. A right subcostal Kocher incision was made, the bile duct was identified and a choledochotomy performed, a stone of approximately 0.9  $\times$  0.9 cm was extracted (Figures 2 and 3) and a T-tube placed to drain the bile. After the procedure, the patient was transferred to the ICU for the management of septic shock, where she



**Figure 3:** *Calculus extracted from common bile duct, with approximate dimensions of 0.9  $\times$  0.9 cm.*

improved and her laboratory returned to almost normal. The multiple abscesses, were treated with antibiotics, metronidazole (500 mg every 8 hours) plus ceftriaxone (1 g every 12 hours) for six weeks. Blood cultures were taken from both arms before antibiotic treatment, which reported no growth. After ten days, the patient was successfully discharged home.

## DISCUSSION

Thrombosis associated with bile duct infection has been little described as the cause of pylephlebitis and represents 7%.<sup>3</sup>

The symptoms of our patient were very nonspecific; general malaise, fever, and nausea.<sup>3</sup> However, the clinical signs were: abdominal pain in the right upper quadrant, hepatomegaly, and jaundice, more associated with pylephlebitis due to cholangitis or liver abscesses.<sup>11</sup> Jaundice is rare in pylephlebitis, and it is associated with advanced forms and significant liver damage.<sup>2</sup> Splenomegaly is found in 10 to 25% of cases.<sup>5</sup>

Leukocytosis is common, although a normal white blood cell count or leukopenia may be found. In this case, a marked leukocytosis was observed at the expense of polynuclear cells. Liver function tests are altered, especially alkaline phosphatase or gamma-glutamyl transferase can multiply their normal value by several times. Jaundice is associated with an increase of up to six times the concentration of bilirubin, and the formation of abscesses contributes to liver dysfunction and coagulation disorders.<sup>3,5,8</sup> Abscesses confirmed on computed tomography could be related to the alterations in coagulation and liver dysfunction detected in this patient.

Blood cultures are positive in 23 to 88% of cases,<sup>12</sup> Highlighted are the three main microorganisms described, *E. coli*, *S. viridans*, and *B. fragilis*, although other organisms have been isolated, such as *Aeromonas hydrophila*, *Streptococci*, *Proteus mirabilis*, *Clostridium spp.* and *K. pneumonia*.<sup>5</sup> In our case, no microorganism was isolated.

Once the diagnosis is made, antibiotic treatment should be started immediately, according to the protocols of each hospital, but it must provide coverage against Gram-negative

and anaerobic bacilli, and maintained for four weeks. The management of liver abscesses requires antibiotic therapy for six weeks and percutaneous or surgical drainage of abscesses greater than 3 cm.<sup>2,4,8</sup> In our case, the focus of infection, the bile duct, was controlled, and antibiotic therapy was indicated for six weeks because of the multiple liver abscesses.

It was decided not to anticoagulate since it is controversial. It is debated whether it can only be reserved for thrombosis with radiological progression, suppurative pylephlebitis, intestinal resection after ischemia secondary to thrombosis, or hypercoagulable states.<sup>5</sup>

Surgery is directed to the treatment of the intra-abdominal focus of infection, for example, appendectomy or cholecystectomy.<sup>2,8</sup> Surgical thrombectomy is associated with higher recurrence rates of thrombosis, and therefore, some studies have advised against it.<sup>3</sup>

Intraoperative exploration of the common bile duct is done according to the surgeon's preferences and experience. Open common bile duct examination is more widely available than laparoscopic examination, but it is associated with higher morbidity (12.7%) as compared to laparoscopic examination (6.5%).<sup>13</sup>

## CONCLUSION

Pylephlebitis is an infrequent complication, very varied and nonspecific in its clinical manifestations.

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