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Case report

Acute pylephlebitis secondary to perforated sigmoid diverticulitis: A case report ^{☆,☆☆}

Luqman Wali, MBBS, MRes^{a,*}, Ali Shah, BSc, MBBS^b, Stefan Sleiman, Bsc, MBBS^c, Tord Hogsand, MUDr^d, Sasha Humphries, MBBS^c

^a Medway NHS Foundation Trust, Kent, UK

^b East Suffolk and North Essex NHS Foundation Trust, Essex, UK

^c Maidstone and Tunbridge Wells NHS Trust, Kent, UK

^d Dartford and Gravesham NHS Trust, Kent, UK

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ABSTRACT

Pylephlebitis is defined as the development of portal venous thrombosis secondary to intra-abdominal infection or inflammation. We present the case of a 69-year-old male with pylephlebitis who attended the Emergency Department with nonspecific abdominal pain, rigors and deranged liver function tests. After an initial negative ultrasound study, computed tomography (CT) scanning demonstrated perforated diverticulitis with an associated thrombus within the portal venous system. Prompt imaging coupled with a high index of clinical suspicion helps in identifying this condition early, significantly reducing morbidity and mortality rates. This case emphasizes the importance of careful evaluation of the portal venous system in cases of intra-abdominal sepsis to exclude this rare, and sometimes fatal, condition.

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Introduction

Pylephlebitis is defined as an inflammatory thrombosis of the portal vein and its tributaries due to intraabdominal infection. Commonly associated conditions include: diverticulitis, appendicitis or cholecystitis. Patients normally present with ambiguous abdominal symptoms. However, timely diagnosis

there are significant complications and a high mortality rate if left untreated. Computed tomography (CT) imaging is routinely used to visualize the extent of thrombus, the underlying cause of pylephlebitis, and any sequelae, such as hepatic compromise. The mainstay of treatment is systemic anti-biotic therapy and anti-coagulation, with poorer outcomes if treatment is delayed. Herein, we present a case of perforated diverticulitis causing pylephlebitis.

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* Corresponding author.

E-mail address: l.wali@nhs.net (L. Wali).

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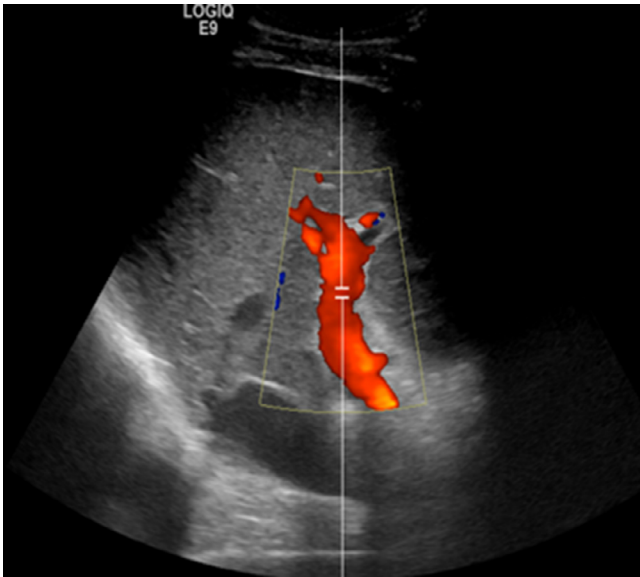


Fig. 1 – Ultrasound study performed shortly after admission demonstrating normal flow within the main portal vein with no intraluminal thrombus.

Case report

A 69-year-old man was admitted to hospital following a 6-day history of dizziness, mild right upper quadrant pain and rigors. He denied any urinary symptoms, nausea, vomiting or respiratory symptoms. Upon admission, he was afebrile with a soft, non-distended abdomen and clear lung fields to auscultation. His blood results showed raised inflammatory markers and mildly deranged liver function tests. His only past medical history of note was of glaucoma, and a recent swab for SARS-CoV-2 returned negative. An initial working diagnosis of ascending cholangitis was made due to his presentation and the cholestatic nature of the abnormal blood results. The patient was initiated on broad spectrum antibiotics pending further investigations.

An ultrasound study of the abdomen was performed the following day which revealed a sonographically normal gallbladder with no gallstones and the absence of any intra- or extra-hepatic biliary dilatation. The liver appeared smooth in contour with regular echotexture and normal flow through the main portal vein was demonstrated (Fig. 1). The appendix, pancreas, spleen and kidneys appeared normal. Essentially, no obvious cause for the deranged liver function tests was identified.

Subsequently, peri-admission blood cultures grew streptococci and bacteroides, suggesting a gastrointestinal source of the causative pathogens. Further blood results also demonstrated worsening inflammatory markers and liver function tests. Due to the discordance between the imaging findings and clinical scenario, a contrast-enhanced CT study of the abdomen and pelvis was performed later in the week (Fig. 2). This revealed a normal biliary tree with no evidence of cholangitis.

There was, however, acute diverticulitis of the sigmoid colon with subtle tiny locules of extramural gas suggesting a localized perforation without abscess formation. Associated inflammatory soft-tissue stranding of the peritoneal reflection, the adjacent sigmoid mesentery, and within the left iliac fossa was noted. In addition, linear intraluminal filling defects consistent with thrombus were seen in the inferior mesenteric vein along its course from the sigmoid colon to its confluence with the splenic vein. Further thrombi and associated locules of intraluminal air were identified within the main portal vein and splenic vein. These appearances were suggestive of acute pylephlebitis secondary to an acutely perforated sigmoid diverticulitis.

The perforated sigmoid colon and concurrent infection were treated conservatively with antibiotics and supportive care without the need for surgical intervention. Due to the multifocal thrombi, the patient was started on treatment-dose dalteparin and a hematology review advised long-term anticoagulation. Furthermore, janus kinase 2 (JAK2) mutation and antiphospholipid screens were ordered to identify any underlying causes for the multiple thrombi. Inflammatory markers and liver function tests improved over the following few days and the patient was discharged with a hematology clinic review booked for a later date.

Discussion

Pylephlebitis is a rare complication of intra-abdominal infection and is defined as a suppurative and inflammatory thrombosis of the portal venous system [1]. It is associated with a high mortality rate of 25% and, unfortunately, diagnosis is often missed due to its nonspecific presentation [2]. Typically, patients present with vague abdominal pain, nausea and pyrexia before advanced sequelae from hepatobiliary involvement, such as hepatomegaly and jaundice, become evident. Incidental radiological diagnosis following an ambiguous clinical presentation is typical of the condition.

Initially, there is localized septic thrombophlebitis adjacent to an area of inflammation, most commonly diverticulitis, appendicitis, cholecystitis or pancreatitis. Thereafter, there is extension into the mesenteric vein, portal vein and eventually intrahepatic venous systems. In our case, there was also extension into the splenic vein. Inherited coagulopathies have not been implicated in the aetiology of pylephlebitis likely due to a lack of data [3]. Causative pathogens tend to be polymicrobial, with *Bacteroides fragilis* being the most commonly isolated organism [4]. Thus, antimicrobial therapy should have broad-spectrum cover which may narrow following the return of blood culture samples. This was the case with our patient who was initiated on meropenem before it was changed to metronidazole following blood culture results. The role of anticoagulants is less clear, however, and the prevalent theory of aggressive anticoagulation seems to be aimed at preventing thrombus extension, complete occlusion or subsequent venous infarction [2].

Definite diagnosis of pylephlebitis through culture-positive aspiration of the portal vein in the setting of portal thrombo-

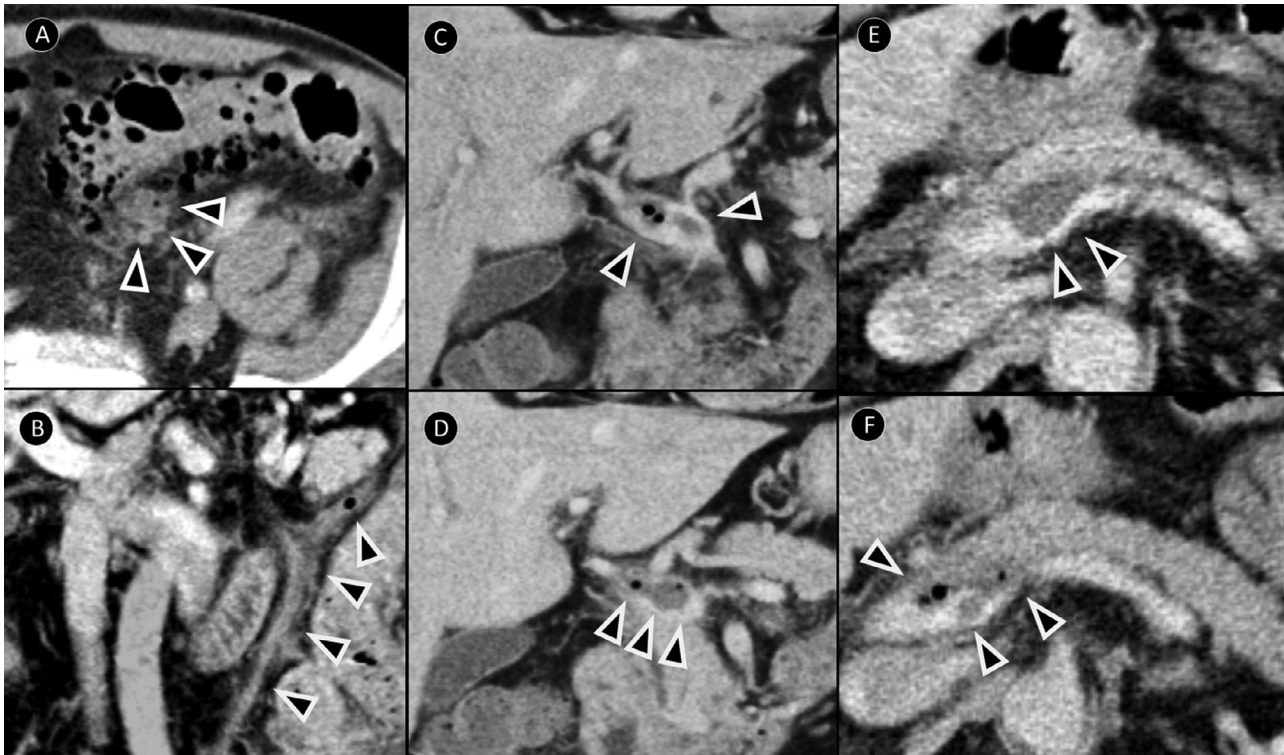


Fig. 2 – Contrast enhanced CT study of the abdomen and pelvis. (A) Axial image of the pelvis showing diffuse sigmoid diverticulitis with inflammatory phlegmon formation and locules of free air (arrowheads). (B) Coronal image demonstrating a thickened and hypoattenuating inferior mesenteric vein with intravenous gas (arrowheads). (C, D) Sequential coronal images demonstrating main portal vein thrombus with associated intravenous gas (arrowheads). (E, F) Sequential axial images demonstrating main portal vein thrombus with associated intravenous gas (arrowheads).

sis is challenging and rarely performed. Instead, the patient's clinical status, blood results and imaging investigations are often used as surrogate markers. Ultrasound may demonstrate flow defects or poor compressibility of the portal venous system. However, overlying bowel gas, patient compliance and variances in operator skill levels may impair accurate diagnosis. The ultrasound study for our patient was normal and flow was seen within the main portal vein. The patient may have developed a thrombus at a later stage or it was simply not identified despite being present.

CT is the modality of choice for the identification of pylephlebitis and any associated intra-abdominal pathology [5]. Direct visualization of thrombi or air within the portal venous system indicates the presence of pylephlebitis. However, CT is insensitive in identifying thrombi that are limited to smaller vessels [6]. Evaluation of the intrahepatic portal venous system and hepatic parenchyma is also essential. Unopacified portal vein branches, intrahepatic abscess formation and parenchymal attenuation differences are all known associations [1,4].

Thrombi in the portal and/or mesenteric veins are found in a number of conditions, including, but not limited to: hematological disorders (eg essential thrombocythaemia, polycythaemia), hypercoagulable states (eg pregnancy, malignancy) and abdominal trauma [6]. A thorough clinical his-

tory coupled with the appropriate use of imaging and clinical correlation of findings is key to identifying pylephlebitis. Deranged liver function tests of unknown origin coupled with nonspecific abdominal pain should warrant further diagnostic imaging with a high index of suspicion for pylephlebitis if more common pathologies seem unlikely. Early detection and aggressive management is key and has led to significantly reduced mortality rates in recent times [2].

Conclusion

Pylephlebitis is a suppurative, ascending infection with thrombosis of the portal venous system that is associated with significant morbidity and mortality. Early detection and aggressive management with antibiotics and anticoagulants can both prevent disease progression and lead to resolution. Therefore, there should be a high index of suspicion for pylephlebitis in patients with intra-abdominal sepsis of unknown source, nonspecific abdominal symptoms and deranged liver function tests. Appropriate imaging, with particular attention to the portal venous system, is key to diagnosis and subsequent management.

Patient consent

The authors obtained written informed consent prior to submission.

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