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Case Report

Pylephlebitis with Bacteremia from a Rare Bacterium: A Case Report of Septic Mesenteric-Portal Thrombophlebitis Associated with Acute Diverticulitis

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Abstract: Pylephlebitis, defined as septic thrombophlebitis of the portal vein, is a rare complication of intra-abdominal infections, particularly appendicitis and diverticulitis. Prompt diagnosis through laboratory tests and imaging, and the early institution of antibiotic therapy, with or without anticoagulation, considerably improve the prognosis. The authors present an atypical case of pylephlebitis associated with uncomplicated acute diverticulitis, with the isolation of a rare bacterium in peripheral blood, *Sutterella wadsworthensis*.

Keywords: Colonic Diverticulitis; Bacteremia; Thrombophlebitis; Venous Thrombosis.

1. Introduction

Pylephlebitis, initially described by William Osler in 1882, is defined as septic thrombophlebitis of the portal vein, a rare complication resulting from intra-abdominal or pelvic infections, with high morbidity and mortality [1-8]. Due to its low incidence, there is a scarcity of prospective, controlled, and randomized studies in the literature to guide its management. For this same reason, the epidemiology of this condition is still uncertain [1, 2, 9, 10]. In February 2023, a systematic review gathered various reports, totaling 220 patients with this diagnosis between 1970 and 2022, suggesting an average age of 50 years and a mortality rate of 14% [2]. Before the antibiotic era, it was considered a fatal disease [6]. Although any intra-abdominal or pelvic infection can cause pylephlebitis, appendicitis and diverticulitis are the most common etiologies [3-5, 11-13].

Septic thrombophlebitis of the portal vein can manifest through sepsis of abdominal origin, presenting signs of systemic inflammation, tachycardia, fever, and hemodynamic and laboratory changes. In the context of acute sigmoid colon diverticulitis, one of the most common etiologies associated with pylephlebitis, the typical clinical findings are localized abdominal pain in the left iliac fossa, possibly accompanied by signs of localized peritonitis, guarding on palpation, changes in bowel habits, fever, and leukocytosis [14]. The portal vein originates from the junction of the superior mesenteric vein and splenic vein, responsible for draining a large part of the intra-abdominal portions of the gastrointestinal tract.

Pylephlebitis arises from thrombophlebitis of small-caliber veins that drain the infection site, extending or migrating to the portal vein [1, 2, 5, 9]. Involvement of mesenteric veins has a higher potential for intestinal ischemia [3, 10]. It is a polymicrobial disease, but in the presence of bacteremia, the most commonly isolated pathogens in peripheral blood are *Bacteroides spp, Escherichia coli*, and *Streptococcus spp* [1,2,10].

The symptoms are usually nonspecific, with most patients presenting with fever and abdominal pain, potentially experiencing nausea, vomiting, and, more rarely, jaundice [1, 6, 13]. Most diagnoses are made after findings in complementary imaging exams, without a high degree of suspicion during initial evaluation [1, 15]. The investigation should include laboratory tests, including a complete blood count, liver function tests, and blood cultures [5, 6, 9, 16], as well as imaging exams such as computed tomography and Doppler ultrasound, with the possibility of using magnetic resonance imaging or PET-CT [1, 6, 17]. Demonstration of portal vein thrombosis is the central finding for diagnosing pylephlebitis. Computed tomography is the exam of choice as it demonstrates not only the thrombosis but also the infection's topography and other possible complications [1, 15]. The most frequent laboratory findings are leukocytosis and high levels of C-reactive protein, with possible elevation of transaminases, canalicular enzymes, and hyperbilirubinemia [9, 18-21].

The treatment consists of antibiotic therapy, initially administered parenterally, with a possible transition to oral administration, typically lasting 4 to 6 weeks [1, 2, 5, 6]. In patients with extensive thrombosis, including mesenteric veins, anticoagulation should be considered [9, 16]. The combination of antibiotic therapy with anticoagulation appears to result in more favorable outcomes [9, 10, 22]. Early diagnosis and treatment substantially improve the prognosis [1]. Multidisciplinary management in treating pylephlebitis is essential, involving not only the general surgeon and gastroenterologist but also assisting with antibiotic therapy and anticoagulation management [1].

This work aims to report the diagnosis and management of a rare case of pylephlebitis, a septic thrombophlebitis of the portal vein, associated with acute sigmoid colon diverticulitis.

2. Case Report

Male patient, 63 years old, admitted to a tertiary hospital for evaluation by the emergency general surgery team, presenting with hypogastric abdominal pain and fever (39°C) in a single episode four days prior to admission, with partial improvement after antibiotic therapy prescribed in an urgent care unit. The patient developed jaundice, prompting him to seek medical attention again, and he was referred to the reference service. Previously healthy, ex-smoker, with no history of surgeries. On physical examination, the patient had stable vital signs, adequate peripheral perfusion, mild jaundice of the sclera and skin, a globose and normotympanic abdomen with present bowel sounds, non-tender on palpation, and no signs of peritonitis. No particularities were noted in the system review. Laboratory tests were performed, and an abdominal and pelvic computed tomography (CT) scan was requested for the differential diagnosis of inflammatory acute abdomen and cholestatic syndrome. Given the jaundice, along with a recent history of abdominal pain and fever, cholangitis and acute diverticulitis were considered as hypotheses.

Laboratory tests showed leukocytosis with a left shift, thrombocytopenia, elevated transaminases, bilirubins, canalicular enzymes, and C-reactive protein, without other alterations (Table 1). The CT scan revealed an area of fat stranding and thin fluid layers in the left iliac fossa around sigmoid colon diverticula (Figure 1), as well as an extensive intraluminal filling defect involving the sigmoid vein (which drains directly to this area), superior rectal veins, the entire extent of the inferior mesenteric vein (Figure 2), the splenomesenteric junction, and the proximal portion of the portal vein (Figure 3), without direct or indirect signs of acute cholangitis, suggesting a diagnosis of thrombophlebitis secondary to sigmoid diverticulitis. The hypothesis of cholangitis was discarded as there were no signs of obstructive factors or dilation of intra- or extra-hepatic bile ducts.

With the assistance of the Hospital Infection Control Committee (HICC) team, blood cultures were collected, and empirical parenteral antibiotic therapy with ceftriaxone and metronidazole was initiated. Due to thrombocytopenia, it was decided to wait for platelet levels to rise above 50,000/mm³ before starting anticoagulation with heparin, which was initiated after two days. Six days after the collection of peripheral blood cultures, the bacterium *Sutterella wadsworthen*sis was isolated; however, an antibiogram was not performed due to the lack of specific sensitivity cut-off patterns for this pathogen due to its low prevalence. The previously initiated antibiotic therapy regimen was maintained under the guidance of the HICC.

Figure 1. Area of fat stranding and thin fluid layers in the left iliac fossa around the diverticula of the sigmoid colon.

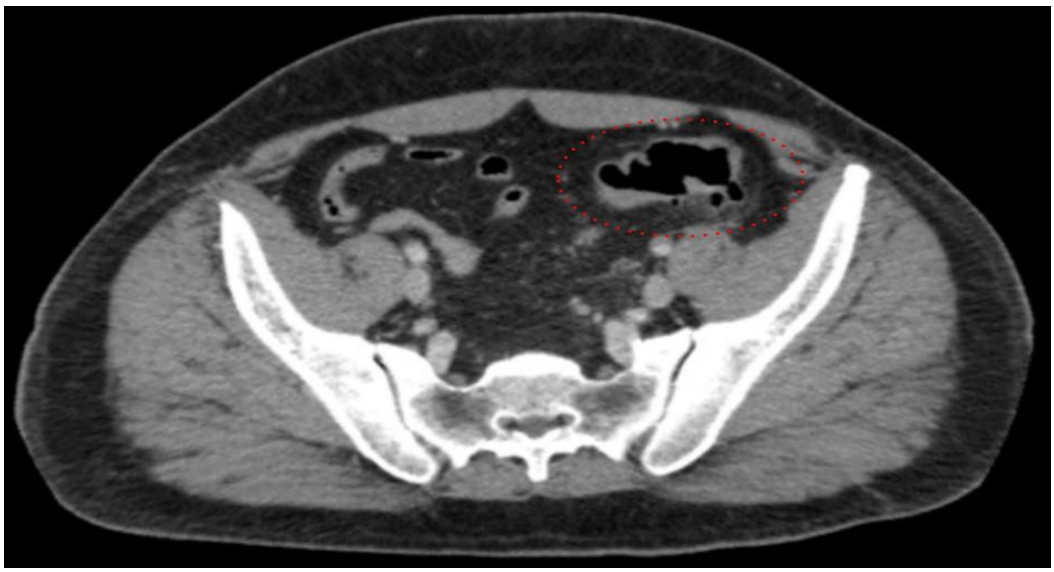


Table 1. Hospital Admission Laboratory Tests.

Parameter	Value	Parameter	Value
Hemoglobin	16.4	Indirect Bilirubin	1.12
Leukocyte Count	11.9	Direct Bilirubin	2.46
Bands	14%	Aspartate Aminotransferase (AST)	102
Platelets	40.9	Alanine Aminotransferase (ALT)	155
Urea	58	Gamma-Glutamyl Transferase (GGT)	616
Creatinine	0.98	Alkaline Phosphatase (ALP)	271
Sodium	135	Amylase	29
Potassium	4.3	Lipase	15
Total Bilirubin	3.58	C-Reactive Protein (CRP)	17.4

Table 2. Laboratory Tests During Hospitalization Until Discharge.

Parameter	Day 1	Day 2	Day 3	Day 4	Day 5	Day 6	Day 7
Hemoglobin (g/dL)	16.4	15.7	14.8	14.3	13.5	13	13.5
Leukocyte Count (/mm³)	11.9	11.8	24.8	23.5	23.4	14	12.7
Bands (%)	14	13	9	11	9	0	0
Platelets (/mm³)	40.9	44.3	55.9	81.6	81.2	255	291

C-Reactive Protein (mg/dL)	17.4	12.19	12.19	10.51	9.52	-	1.47
Total Bilirubin (mg/dL)	3.58	3.8	5.19	4.32	3.46	-	-
Direct Bilirubin (mg/dL)	2.46	2.49	3.81	3.09	2.31	-	-

Figure 2. Stranding with intraluminal filling defect involving the sigmoid vein (which drains directly to this area), superior rectal veins, and the entire extent of the inferior mesenteric vein.

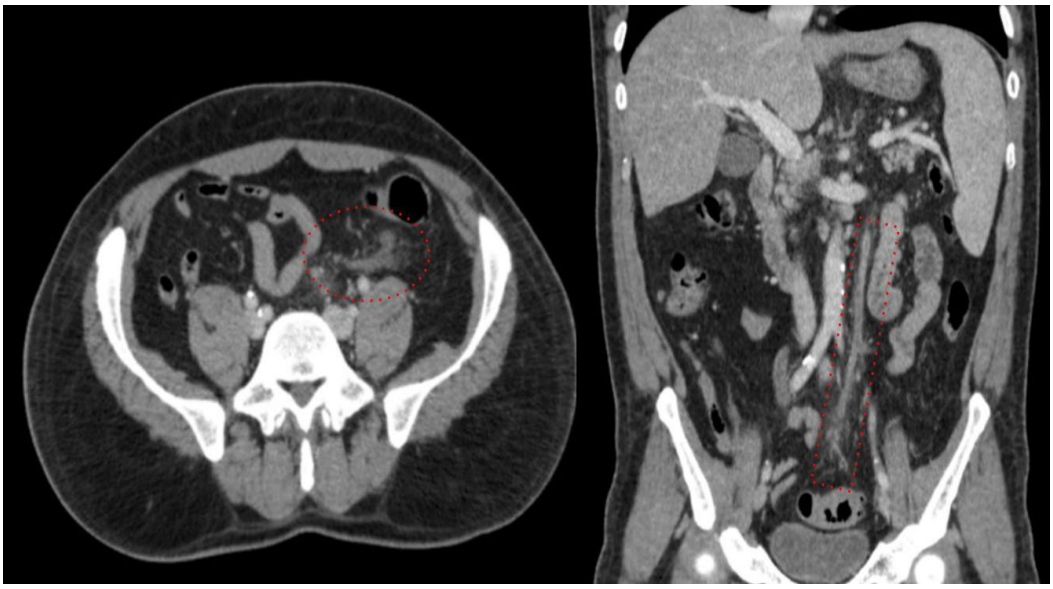
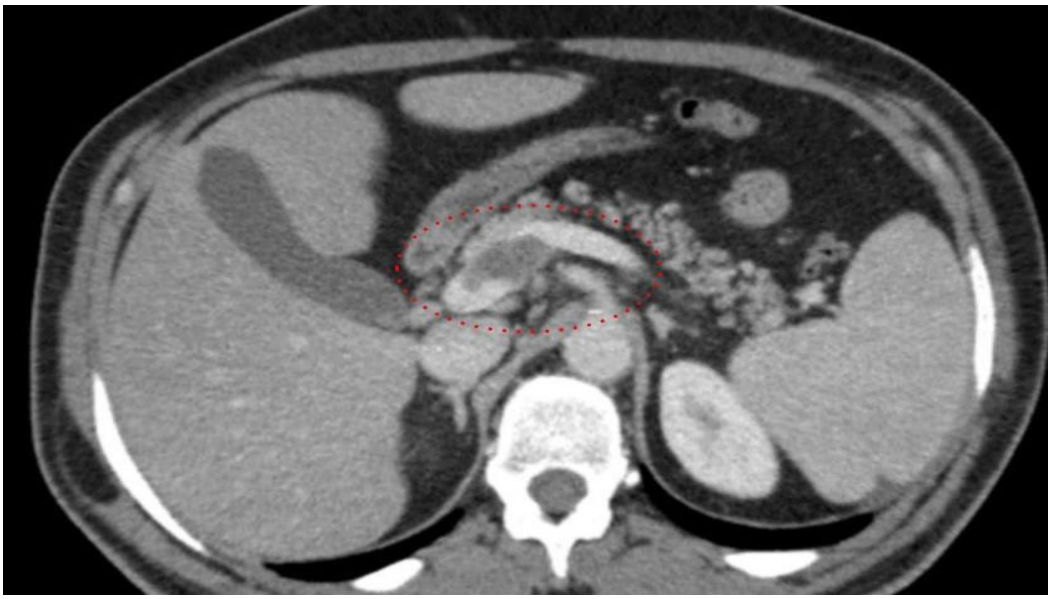


Figure 3. Filling defect at the splenomesenteric junction and proximal portion of the portal vein, compatible with thrombosis.



S. wadsworthensis is a gram-negative bacterium belonging to the genus *Sutterella*. The treatment of septic thrombophlebitis associated with this pathogen presents significant challenges due to the rarity of this bacterium and the lack of specific antimicrobial sensitivity data. Without a targeted antibiogram, the empirical choice of antimicrobials must be based on clinical knowledge, considering the intrinsic resistance of *Sutterella* and the

need to avoid excessive use of antibiotics. The choice of ceftriaxone is based on its effectiveness against gastrointestinal tract infections and its penetration into the central nervous system. Metronidazole is an antimicrobial with activity against anaerobic bacteria, including many intestinal species such as *S. wadsworthensis*.

The patient showed substantial improvement in leukocytosis and inflammatory laboratory markers, as well as liver function tests (Table 2), remaining asymptomatic and clinically stable, with no abdominal pain, fever episodes, or hemodynamic instability. After 7 days of hospitalization, the patient was discharged in good condition with oral antibiotic therapy of Ciprofloxacin 500 mg twice a day and Metronidazole 500 mg three times a day, aiming to complete four weeks of treatment, along with anticoagulation with Apixaban 5 mg every 12 hours, with a plan for early outpatient reassessment with imaging, in conjunction with the vascular surgery and coloproctology teams.

At the initial outpatient follow-up and after one month, the patient was completely asymptomatic, non-icteric, with regular bowel habits and a non-tender abdomen. Laboratory tests were within normal limits, with no altered infectious parameters, thrombocytopenia, or hyperbilirubinemia. A follow-up CT scan performed after eight weeks of treatment showed diverticulosis in the sigmoid colon, with no findings suggestive of acute diverticulitis. Additionally, there was resorption of the previously present extensive thrombus, allowing the discontinuation of anticoagulation. During follow-up with coloproctology, the patient is scheduled to undergo a colonoscopy to evaluate the diverticular disease of the colon and screen for neoplasms.

3. Discussion

For the proper diagnosis of pylephlebitis, the use of imaging exams, especially computed tomography, is indispensable [1, 6, 15, 17]. Timely identification and treatment can change the natural course and progression of this disease, preventing major complications such as intestinal perforation consequent to diverticulitis and intestinal ischemia due to extensive venous thrombosis, as well as death in cases of late diagnosis [1-8, 10].

Antibiotic therapy should be instituted in all cases, ideally guided by culture exams or empirically to cover the most prevalent pathogens [1,2,5-7]. Identifying the involved pathogen for better study and guidance for antibiotic therapy through blood culture collection is extremely important. Besides the most common microorganisms isolated in peripheral blood, there are also reports in the literature of microorganisms such as *Aeromonas hydrophila, streptococci, Proteus mirabilis, K. pneumoniae, anaerobic streptococci, Clostridium spp, Fusobacterium, Parvimonas*, and *Actinomyces* [23-26]. In a series of three cases, Kirk and colleagues described the identification of the bacterium *Sutterella wadsworthensis* in blood cultures of patients who underwent abdominal surgeries during acute abdomen (complicated diverticulitis and appendicitis) [27]. There are no previous reports in the literature of *S. wadsworthensis* isolation during pylephlebitis.

Sutterella wadsworthensis is a rare Gram-negative microaerophilic bacterium associated with intestinal inflammatory conditions such as ulcerative colitis, Crohn's disease, acute diverticulitis, and appendicitis. Despite this, there is still no complete elucidation of its pathophysiological predilection for the gastrointestinal tract. The infection occurs after the pathogen invades the cell membrane of the intestinal wall and disseminates hematogenously, potentially leading to infected portal vein thrombosis and, consequently, pylephlebitis. Its clinical implications involve chronic intestinal inflammation, perforation, abscess, and obstruction. However, the isolated clinical consequence of this bacterium cannot yet be measured in the scientific literature due to the scarcity of identified cases and the limited association with other infectious agents causing pylephlebitis [28-31].

The need for anticoagulant therapy and its specific regimen should be evaluated individually, with an apparent important role in preventing intestinal ischemia, resulting in positive outcomes in reducing morbidity and mortality and hospitalization of patients with pylephlebitis [9, 16, 18, 22, 32-36]. In refractory cases with progression of mesenteric

vein thrombosis at risk of ischemia, thrombolytic therapy may be an option, and in established ischemia, surgical resection with evaluation of intestinal viability is necessary [33, 36]. According to the *American Association for the Study of Liver Diseases* guidelines, although limited, there are retrospective data suggesting that anticoagulation, concomitant with antibiotic therapy, in the treatment of these patients, results in higher rates of complete resolution and fewer long-term portal hypertension complications [37].

4. Conclusion

In this article, we present a case report of an elderly male patient diagnosed with septic pylephlebitis. This condition is a rare complication associated with intra-abdominal infection and, in this specific case, related to uncomplicated acute sigmoid colon diverticulitis. The highlight of this case also lies in the isolation of an atypical pathogen in peripheral blood cultures: *Sutterella wadsworthensis*. It is important to note that *Sutterella wadsworthensis* is rarely described in the medical literature, and until now, no previous reports have linked this microorganism to pylephlebitis. The uniqueness of this case reinforces the need to consider this condition in patients with abdominal symptoms and suspected infection, especially when microbiological results point to an uncommon agent.

We highlight the successful resolution of the infection and thrombophlebitis after treatment with antibiotic therapy and anticoagulation. The rapid identification and early management of pylephlebitis are crucial for patient survival. In summary, our experience illustrates the importance of early diagnosis and appropriate treatment, positively impacting the clinical evolution and quality of life of patients. Pylephlebitis, once considered fatal, can be successfully managed when approached diligently and informed.

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Conflicts of Interest: None.

Supplementary Materials: None.

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