Intest Res 2009;7(2):105-109

하부장간막정맥과 문맥의 문맥염 1예

이화여자대학교 의학전문대학원 내과학교실, 의과학연구소

정해선 · 심기남 · 정지민 · 강민정 · 나윤주 · 정성애 · 유 권

A Case of Pylephlebitis of the Inferior Mesenteric Vein and Portal Vein

Hae Sun Jung, M.D., **Ki-Nam Shim,** M.D., **Ji Min Jung,** M.D., **Min-Jung Kang,** M.D., **Youn Ju Na,** M.D., **Sung Ae Jung,** M.D., **Kwon Yoo,** M.D.

Department of Internal Medicine, Ewha Medical Research Institute, Ewha Womans University School of Medicine, Seoul, Korea

Pylephlebitis is defined as septic thrombophlebitis of the portal vein or one of its tributaries. Pylephlebitis is an uncommon and often fatal complication of intra-abdominal infections, such as diverticulitis and appendicitis. The most common bacteria isolated from patients with pylephlebitis are *Escherichia coli* and *Bacteroides fragilis*. The overall mortality rate is 32%. We describe a case of septic thrombophlebitis of the main portal vein and inferior mesenteric vein successfully treated with broad-spectrum antibiotics and anticoagulants. The early diagnosis and treatment with the timely administration of antibiotics is most important for pylephlebitis. **(Intest Res 2009;7:105-109)**

Key Words: Pylephlebitis; Cavernous Sinus Thrombosis; Inferior Mesenteric Vein; Portal Vein

INTRODUCTION

Pylephlebitis or septic thrombophlebitis of the portal vein (PV) and its tributaries is a serious condition with significant morbidity and mortality.[1] Pylephlebitis can complicate any intra-abdominal or pelvic infection, such as diverticulitis, appendicitis, and cholangitis. The inciting focus is a hypercoagulable state associated with malignancy or clotting factor deficiencies.[1,2] The most common bloodstream isolates from patients with pylephlebitis are *Bacteroides fragilis*, *Escherichia coli*, *Proteus mirabilis*, and aerobic gram-negative bacilli.[1,2] Pylephlebitis is diagnosed by demonstration of portal vein thrombosis accompanied by bacteremia in a febrile patient. CT scan and color flow Doppler ultrasonography are diagnostic.[3] Pylephlebitis can be treated with antibiotics and eradication of the septic focus. We present a case of septic thrombophlebitis of the main PV and inferior mesenteric vein (IMV) in a 49-year-old man.

CASE REPORT

A 49-year-old man presented with a 7-day history of fever, left flank pain, and diarrhea. He had no other significant medical history. He did not take any medications. On admission, the abdominal examination revealed left upper quadrant tenderness without rebound tenderness. On examination, his temperature was 37.9°C, the blood pressure was 105/70 mmHg, and the heart rate was 92 beats/minute.

접수 : 2009년 3월 23일 수정 : 2009년 8월 26일
승인 : 2009년 10월 29일
• 연락처 : 심기남, 서울시 양천구 목동 911-1 (157-710)
이화여자대학교 의과대학 부속목동병원 소화기내과
Tel: 02) 2650-2632, Fax: 02) 2655-2076
E-mail: shimkn@ewha.ac.kr

Received : March 23, 2009. Revised : August 26, 2009.
Accepted : October 29, 2009.
• Correspondence to : Ki-Nam Shim, M.D., Department of Internal Medicine, Ewha Womans University School of Medicine, 911-1, Mok-dong, Yangcheon-gu, Seoul 157-710, Korea
Tel: +82-2-2650-2632, Fax: +82-2-2655-2076
E-mail: shimkn@ewha.ac.kr

The laboratory data showed the following: hemoglobin, 11.8 g/dL; hematocrit 33.4%; and a white blood cell (WBC) count, 22,500/mm^3 with 79% segmented neutrophils and a platelet count of 383,000/mm^3. The erythrocyte sedimentation rate (ESR) was 25 mm/hr and the C-reactive protein was 9.7 mg/dL. The prothrombin time and partial prothrombin time were 11.5 seconds, with an international Normalized Ratio (INR) of 1.07 and 23.8 seconds. Liver function tests revealed the following values: alkaline phosphatase, 388 IU/L; γ-glutamyltransferase, 330 IU/L; alanine aminotransferase, 27 IU/L; aspartate aminotransferase, 38 IU/L; total bilirubin, 1.3 mg/dL; and direct bilirubin, 0.9 mg/dL. Other laboratory data showed no abnormal findings. The results of chest and abdominal simple X-ray were normal and the electrocardiogram showed sinus tachycardia (115 beats/minute). A CT scan of the abdomen and pelvis showed enhanced wall thickening with an intraluminal filling defect in the IMV and the main PV, a dilated lumen of the IMV, and soft tissue inflammation surrounding the IMV. Also, a CT scan showed diverticulitis at the sigmoid colon. Therefore, a CT scan demonstrated evidence of acute thrombophlebitis within the IMV and the main PV with diverticulitis (Fig. 1). He was treated empirically with broad-spectrum antibiotics (ceftriaxone (2 g per day) and metronidazole (500 mg qid)), and he was fully anticoagulated with subcutaneous enoxaparin sodium. He had an isolated high spiking fever the day after admission, after which the fever subsided. Left flank pain and diarrhea was improved on the second hospital day. Seven days later, *Bacteroides fragilis* was recovered from his blood from admission. To find the other cause of thrombophlebitis, a radiologic and endoscopic examination was performed. An ultrasound of the lower extremity demonstrated no deep vein thrombosis and the echocardiography was normal. An upper endoscopy showed raised erosive gastritis and a duodenal ulcer scar. A colonoscopy showed a colon polyp. A small

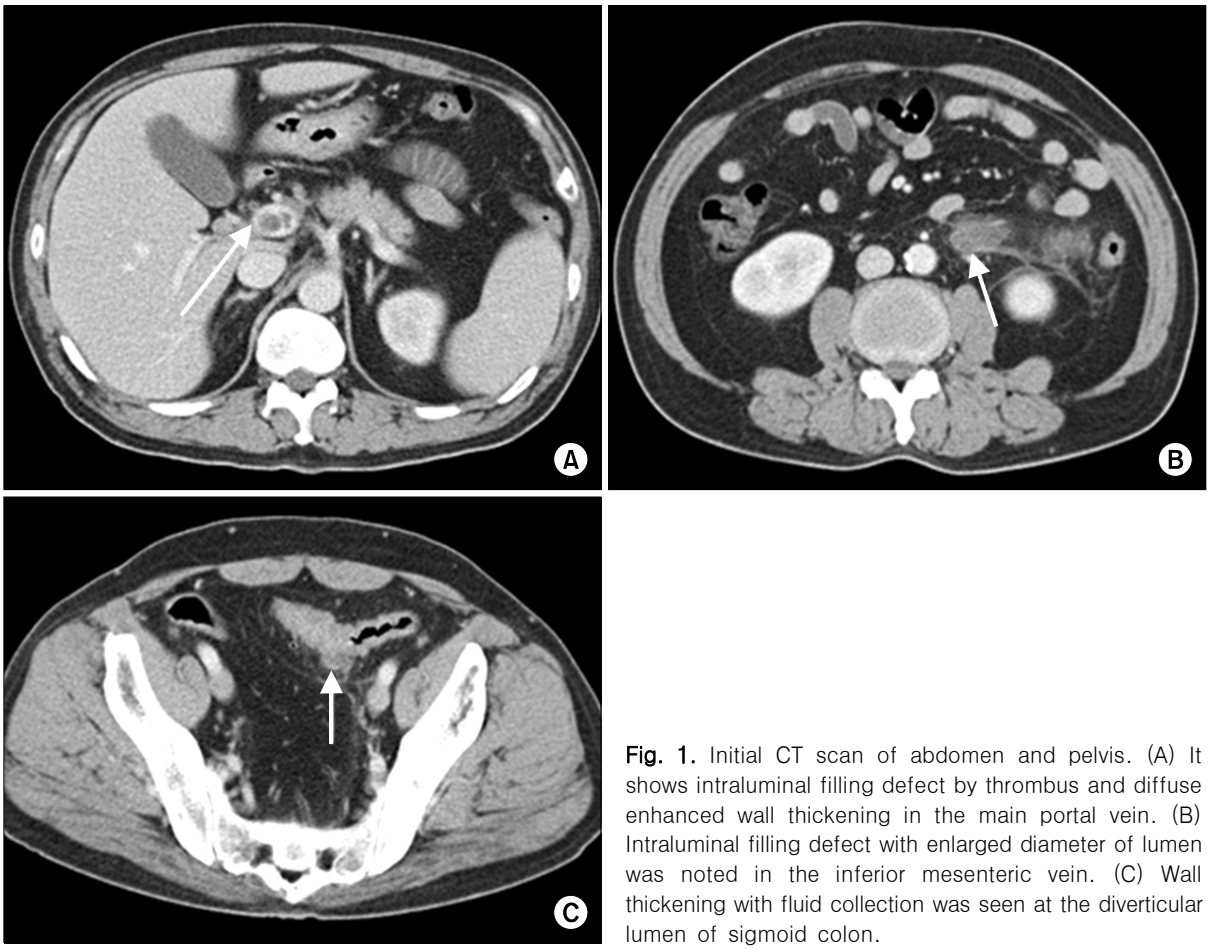

Fig. 1. Initial CT scan of abdomen and pelvis. (A) It shows intraluminal filling defect by thrombus and diffuse enhanced wall thickening in the main portal vein. (B) Intraluminal filling defect with enlarged diameter of lumen was noted in the inferior mesenteric vein. (C) Wall thickening with fluid collection was seen at the diverticular lumen of sigmoid colon.

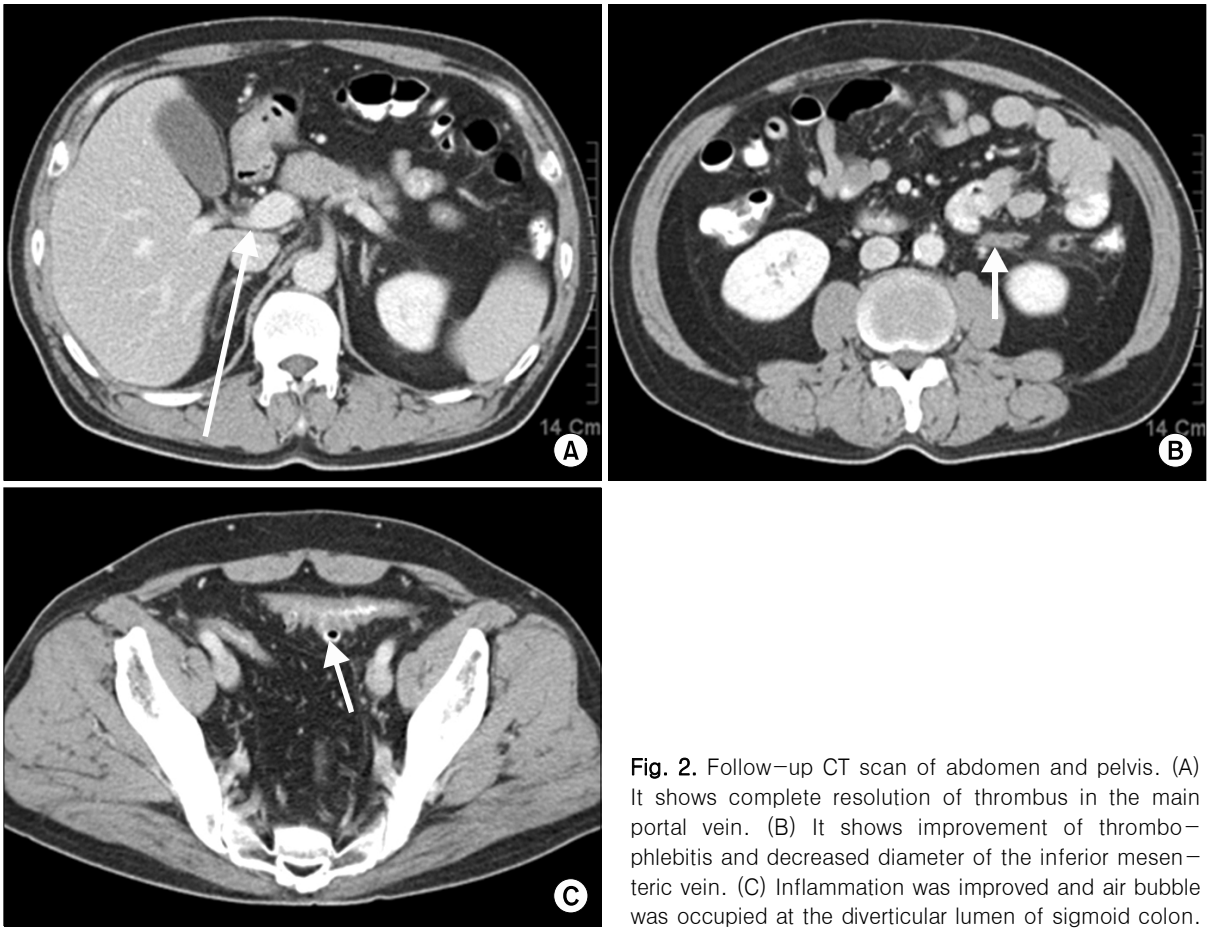

Fig. 2. Follow-up CT scan of abdomen and pelvis. (A) It shows complete resolution of thrombus in the main portal vein. (B) It shows improvement of thrombophlebitis and decreased diameter of the inferior mesenteric vein. (C) Inflammation was improved and air bubble was occupied at the diverticular lumen of sigmoid colon.

bowel examination showed diverticuli at the 2nd and 3rd portions of the duodenum. He recovered uneventfully and was discharged after 9 days. He had oral antibiotics for 7 days and enoxaparin sodium for 2 months after discharge. A repeat CT scan of the abdomen and pelvis performed after 1 month of discharge revealed improvement in the thrombophlebitis, disappearance of the thrombus in the IMV and the main PV, and improvement of diverticulitis of the sigmoid colon (Fig. 2).

DISCUSSION

Pylephlebitis is defined as septic thrombophlebitis of the PV or one of its tributaries.[1] Pylephlebitis is a rare complication of an intra-abdominal inflammatory process and a serious condition with significant morbidity and mortality that can complicate intra-abdominal sepsis. The mortality rate of pylephlebitis ranges from 11-32%.[1,3]

The most common cause of pylephlebitis is diverticulitis.[1] Although diverticulitis is the most common cause of pylephlebitis, it remains a rare complication of diverticulitis.[2] In one report by Kaewlai,[4] only 1 of 138 patients with diverticulitis complicated with pylephlebitis. Other causes of pylephlebitis are appendicitis, cholangitis, inflammatory bowel disease (IBD), pyogenic pancreatitis, intestinal perforation, malignancy, and coagulation factor deficiencies.[1,3,5] Among the causes, pylephlebitis in IBD patients can be caused by coagulation abnormalities induced by inflammation or steroid therapy and an immunosuppressive effect secondary to steroid therapy. In IBD patients, subsequent surgical resection of the affected ileum is required after medical treatment.[6,7]

The clinical features of pylephlebitis are non-specific. Fever, chills, abdominal tenderness, and leukocytosis are the initial clinical findings. Jaundice is infrequently present and occurs late in extensive hepatic involvement, such as liver

abscess.[1,8] Clinical features depend on the site of the primary inflammatory process and the severity of the associated infection. The high mortality and morbidity of pylephlebitis arises from the non-specificity of the symptoms and signs.

Bacteremia is frequently present, and positive blood cultures are obtained in approximately 80% of blood samples.[1] The most common isolated bacteria are *Escherichia coli* and *Bacteroides fragilis*. Other isolated organisms include *Proteus mirabilis*, *Streptococcus* species, and aerobic gram-negative bacilli. *Enterococci* are uncommon.[1,3,8,9] In our case, *Bacteroides fragilis* was recovered from the blood.

Ultrasonography can demonstrate the progression or extension of the thrombus within the PV; however, an abdominal CT scan is preferred because ultrasonography is operator- dependent. An abdominal CT scan is potentially diagnostic in the evaluation of portal and mesenteric vein thromboses, and may also identify the primary source of infection.[10,11]

The primary treatment of pylephlebitis is broad-spectrum antibiotic therapy and eradication of the underlying septic focus. Empirical antibiotic therapy should include broad coverage for enteric facultative gram-negative bacilli and anaerobes, especially *B. fragilis* and *Streptococcus* species. Even in the absence of an intra-abdominal source, broad spectrum antibiotics covered with gut-associated organisms should be considered.[12]

The role of anticoagulation is unclear.[1] Currently, many authors consider heparin treatment to be essential for stopping the progression of the thrombosis and eliminating the source of septic emboli.[13-15] A retrospective review of 44 patients with pylephlebitis showed that patients who received anticoagulation had better outcomes than those who did not receive anticoagulation.[3] Anticoagulation could be used in cases of acute extensive pylethrombosis, progression of thrombus after initial diagnosis, and enteric resection for pylethrombosis-induced ischemia.[13] Systemic heparin therapy is initiated with a bolus injection of 5000 U, followed by a continuous infusion in which the dose is adjusted so that the activated partial thromboplastin time remains more than twice the normal level. Oral anticoagulation with warfarin should be started once there is evidence of the absence of ongoing ischemia.[14] The clinical endpoint of anticoagulation therapy in pylephlebitis has not been established. In the absence of ongoing thrombotic disorder, the duration of anticoagulation may be limited to 6 months to 1 year.[14] In our case, after the patient received anticoagulation for 1 month, a follow-up CT scan showed complete improvement of thrombophlebitis and disappearance of thrombus in the PV and IMV without complications. The use of anticoagulation has a complication rate of as high as 20%, and the routine use of anticoagulation must be carefully considered.[16]

Prompt surgical intervention is required only if there is evidence of synchronous liver abscesses, intestinal or mesenteric ischemia, complications of diverticulitis (perforation and bleeding), or when medical conservative therapy fails.[17]

Outcome depends on multiple factors, including age >70 years, the presence of co-existing conditions, concomitant intra- or extra-hepatic abscesses, and the timing of the diagnosis and surgical intervention.[14,18] The high morbidity and mortality of pylephlebitis with hepatic abscesses are attributable to delayed diagnosis and treatment due to non-specific symptoms and signs.[18]

In conclusion, pylephlebitis is an uncommon and often fatal complication of intra-abdominal infection. A high index of suspicion and timely broad-spectrum antibiotics and anticoagulation therapy are required.

REFERENCES

1. Plemmons RM, Dooley DP, Longfield RN. Septic thrombophlebitis of the portal vein (pylephlebitis): diagnosis and management in the modern era. Clin Infect Dis 1995;21: 1114-1120.
2. Saxena R, Adolph M, Ziegler JR, Murphy W, Rutecki GW. Pylephlebitis: a case report and review of outcome in the antibiotic era. Am J Gastroenterol 1996;91:1251-1253.
3. Baril N, Wren S, Radin R, Ralls P, Stain S. The role of anticoagulation in pylephlebitis. Am J Surg 1996;172:449-453.
4. Kaewlai R, Nazinitsky KJ. Acute colonic diverticulitis in a community-based hospital: CT evaluation in 138 patients. Emerg Radiol 2007;13:171-179.
5. Kim TD, Kim TN, Oh HJ, et al. Clinical characteristics and radiologic findings of pylephlebitis. Korean J Med 2005;69: 798-806.
6. Aguas M, Bastida G, Nos P, Beltrán B, Grueso JL, Grueso J. Septic thrombophlebitis of the superior mesenteric vein and multiple liver abscesses in a patient with Crohn's disease at onset. BMC Gastroenterol 2007;7:22.
7. Baca B, Hamzaoğlu I, Karahasanoğlu T, Hamzaoğlu HO. Laparoscopic treatment of pyogenic liver abscess complicating Crohn's disease: a case report. Turk J Gastroenterol 2007;18: 58-61.

8. Navarro C, Clain DJ, Kondlapoodi P. Perforated diverticulum of the terminal ileum. A previously unreported cause of suppurative pylephlebitis and multiple hepatic abscesses. Dig Dis Sci 1984;29:171-177.
9. Perez-Cruet MJ, Grable E, Drapkin MS, Jablons DM, Cano G. Pylephlebitis associated with diverticulitis. South Med J 1993;86:578-580.
10. Balthazar EJ, Gollapudi P. Septic thrombophlebitis of the mesenteric and portal veins: CT imaging. J Comput Assist Tomogr 2000;24:755-760.
11. Lee L, Kang YS, Astromoff N. Septic thrombophlebitis of the inferior mesenteric vein associated with diverticulitis CT diagnosis. Clin Imaging 1996;20:115-117.
12. Wireko M, Berry PA, Brennan J, Aga R. Unrecognized pylephlebitis causing life-threatening septic shock: a case report. World J Gastroenterol 2005;11:614-615.
13. Duffy FJ Jr, Millan MT, Schoetz DJ Jr, Larsen CR. Suppurative pylephlebitis and pylethrombosis: the role of anticoagulation. Am Surg 1995;61:1041-1044.
14. Kumar S, Sarr MG, Kamath PS. Mesenteric venous thrombosis. N Engl J Med 2001;345:1683-1688.
15. Condat B, Pessione F, Helene Denninger M, Hillaire S, Valla D. Recent portal or mesenteric venous thrombosis: increased recognition and frequent recanalization on anticoagulant therapy. Hepatology 2000;32:466-470.
16. Wessler S, Gitel SN. Warfarin. From bedside to bench. N Engl J Med 1984;311:645-652.
17. Nobili C, Uqqeri F, Romano F, et al. Pylephlebitis and mesenteric thrombophlebitis in sigmoid diverticulitis: medical approach, delayed surgery. Dig Liver Dis 2007;39:1088-1090.
18. Lim HE, Cheong HJ, Woo HJ, et al. Pylephlebitis associated with appendicitis. Korean J Internal Med 1999;14:73-76.