

Enterocolic Lymphocytic Phlebitis with Disseminated Intravascular Coagulation-Like Features - A Case Report

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ABSTRACT

We report herein a case of colonic infarction caused by lymphocytic phlebitis. Due to its very rare occurrence, a correct diagnosis was not made until a surgical specimen was obtained. Initially, the patient was treated conservatively as severe colitis, but subsequent laboratory test results suggested worsening of the general condition including disseminated intravascular coagulation (DIC), thus, colectomy was performed. After the operation, the patient's condition improved rapidly and completely. The excessive consumption of platelets and coagulation factors, which promoted the DIC-like features, was thought to have been restricted to the inflamed veins of the damaged colon.

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Key words: Colonic ischemia; Venous thrombosis; Lymphocytic phlebitis; Disseminated intravascular coagulation

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INTRODUCTION

Colonic infarction/ischemic colitis is a group of clinicopathologic entities, which involves various colonic circulatory disorders with different etiopathologies. Enterocolic lymphocytic phlebitis, the pathological mechanism of which has not yet been elucidated, is a rare cause of colonic ischemia^[1-4]. By 2007, only 34 patients with this

illness had been reported in the English literature^[5]. A characteristic of this disorder is lymphocytic infiltration that selectively affects small peripheral branches of the mesenteric vein and induces thrombotic occlusion in the damaged veins. Recently, we successfully treated a patient with colonic infarction that was caused by lymphocytic phlebitis. In this case, however, the treatment strategy was difficult to be made because of the undeterminable etiology and complex clinical course.

CASE PRESENTATION

A 78-year-old housewife who suffered from persistent diarrhea consulted a local clinic. She had no notable medical history other than hypertension and dyslipidemia. The physician suspected ulcerative colitis, and administered 5-aminosalicylic acid (5-ASA). Despite the medication, the diarrhea worsened and melena subsequently developed. Two weeks later, she was referred to our hospital and admitted to the Gastroenterology unit for detailed examinations.

Upon admission, the patient appeared exhausted, and had low-grade fever (37.2°C; Figure 1). Her abdomen was slightly distended but soft, and no tender point was found. Bowel sound was weak. Laboratory test results showed elevated serum level of C-reactive protein (14.14 mg/dL) and moderate thrombocytopenia (67×10^9 count/L; Figure 1). Although no clinical manifestation of coagulopathy was observed, plasma levels of d-dimer and fibrin degradation products were markedly elevated (95.1 mg/L and 167.4 mg/L, respectively). Colonoscopy was performed, and multiple ulcer lesions and erosion were found along the entirety of the colon (Figure 2A). Biopsied mucosal tissue showed erosion and prominent mononuclear cell infiltration (Figure 2B). The attending doctors tentatively diagnosed her illness as severe colitis associated with disseminated intravascular coagulation (DIC), and immediately began administration of antibiotics and protease inhibitors. Computed tomography revealed marked swelling of the colon, particularly in the ascending colon, and small amount of ascites (Figure 3). No thrombotic occlusion was observed in the major mesenteric vessels.

Despite the intensive care, the patient's condition did not improve. Ascites progressively increased, and dyspnea further developed. No pathogenic microorganisms were detected in the fecal and blood specimens. As an alternate treatment strategy, administration of steroids was initiated but the efficacy was not as expected, and thrombocytopenia worsened further (33×10^9 count/L). Although the pathophysiology was still obscure, the necessity of removal of the damaged colon was placed under consideration. On the 12th day of hospitalization, subtotal colectomy and ileostomy were performed. The resected colon showed extensive hemorrhagic infarction, numerous

venous thrombi, and lymphocytic infiltration surrounding the veins (Figure 4 and Figure 5). The final pathologic diagnosis was colonic infarction caused by enterocolic lymphocytic phlebitis. In contrast to the serious preoperative condition, the postoperative recovery was incredibly rapid and straightforward; the circulating platelet count completely recovered (170×10^9 count/L) on the first postoperative day (Figure 1). Five months later, a second operation was carried out to close the stoma, and the patient's condition has since been uneventful.

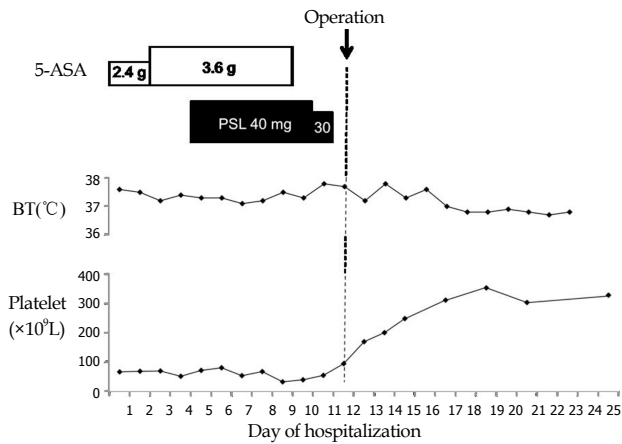


Figure 1 The clinical course during hospitalization. 5-ASA: Aminosalicylic acid; PSL: Prednisolone; BT: Body temperature.

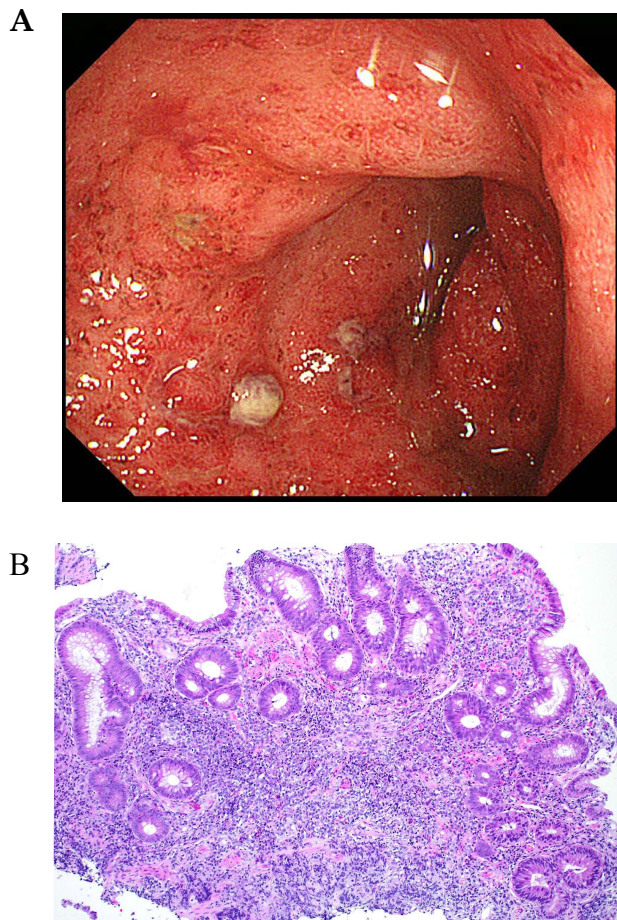


Figure 2 Colonoscopic findings on the first day of hospitalization. A: Multiple ulcers are seen on the reddened and swollen mucous membrane. B: Histologically, damaged mucosa shows erosion and prominent mononuclear cell infiltration.

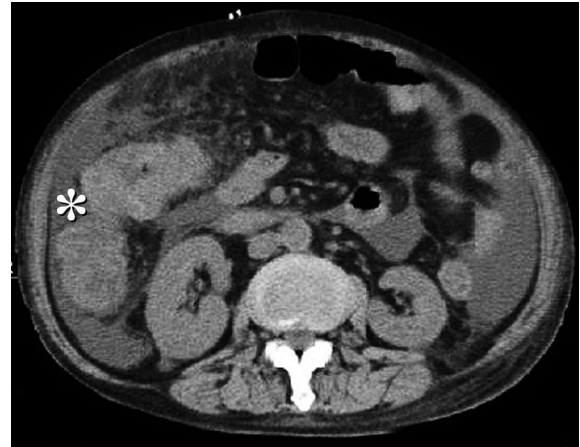


Figure 3 Computed tomography on the tenth day of hospitalization. Swelling of the ascending colon (asterisk) is prominent. Thrombus is undetectable in the large mesenteric vasculatures.

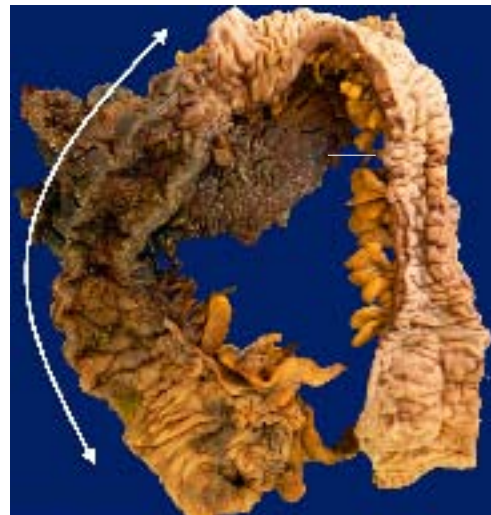


Figure 4 Surgical specimen. The resected colon shows hemorrhagic infarct (an extent indicated with a line with arrow tips) and notable edema.

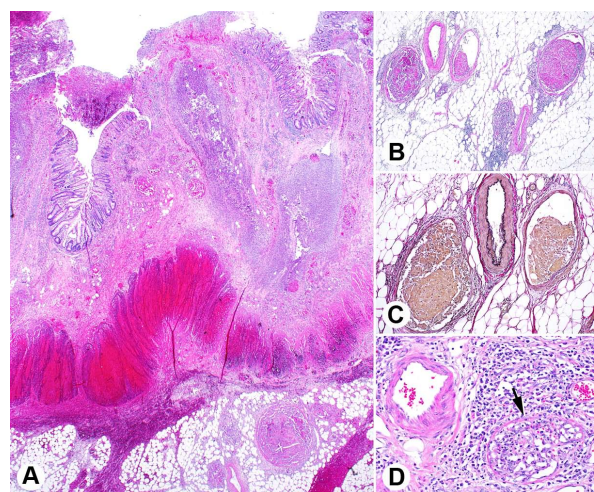


Figure 5 Typical histopathological findings. A: Transmurular necrosis, hemorrhage, dense inflammatory cell infiltration, and multiple thrombi are seen in the damaged colonic tissue (Hematoxylin and eosin, low-magnification); B: Numerous thrombi, either fresh or organizing, are seen in the small mesenteric vasculatures with lymphocytic infiltration. (Hematoxylin and eosin, moderate-magnification); C: A high-magnification view of upper left corner of Figure 5B (Elastica Van Gieson). Note that thrombotic occlusion is limited only to the veins; D: The same selective thrombophlebitis is seen also in a submucosal vessel (arrow; Hematoxylin and eosin, high-magnification).

DISCUSSION

The key aspect of this case was that an early and correct diagnosis was not made, which would have appropriately and promptly averted all the complexities associated with this illness. A clearer insight into the laboratory data that showed DIC-like features was retrospectively thought to have been a clue to reveal the accurate pathophysiology. DIC is a consumptive coagulopathy induced by overstimulation of the coagulation system in various pathological settings^[6]. We initially thought that the DIC resulted from severe colonic inflammation; a catastrophic inflammatory reaction might have systemically evoked such a hypercoagulable status. However, the surgical specimen and postoperative course indicated that consumption of platelets and coagulation factors was limited to the veins of the diseased colon. The colonic damage was an ischemic disorder attributed to venous thrombosis manifesting the DIC-like features in laboratory tests.

We had also considered ischemic disorders as differential diagnoses, but the absence of thrombi in the main trunks of the mesenteric artery and vein excluded them from the initial diagnosis. In addition, the very low incidence of such highly selective colonic venous thrombosis also contributed to our confusion. This rare form of ischemic intestinal disorder due to thrombosis in the colonic veins has been reported with various terminologies in the literature^[1-5]. Preexisting colonic phlebitis is a pathologic basis of this thrombotic circulatory disorder; i.e., the acute ischemic event is caused by chronic and persistent inflammatory vascular damage by lymphocytes^[2]. The next question is “what is a cause of this selective phlebitis which affected only the peripheral branches of the mesenteric vein?” Wright *et al.*^[3] has suggested that colonic mucosal inflammation due to microscopic (lymphocytic) colitis may play a significant role in inducing lymphocytic phlebitis. Early intensive suppression of venous and mucosal inflammation, such as via high-dose steroid administration, may lead to remission of the disease without surgical removal of the affected colon. 5-ASA was not effective in our case. 5-ASA may be insufficient to control the vasculitic process, or even pathogenic in this kind of vasculitis/phlebitis via hypersensitivity reaction^[7]. With the exception of extensive transmural infarction, most colonic ischemic disorders are generally

recognized to be able to be managed conservatively^[8]. Although the outcomes of surgery for this type of severe colonic ischemia are favorable, an alternative, non-surgical treatment is sought to minimize the patient’s burden.

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