Case Report

Phlebosclerotic Colitis: A Case with a History of Herbal Ingestion

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Phlebosclerotic colitis is a rare type of ischemic colitis, characterized by calcification at the right hemicolon. Approximately 40 such cases have been reported, and the patients in most cases were treated surgically. The etiology of phlebosclerotic colitis remains unknown, although portal hypertension is thought to be a cause. The particular radiologic findings of the disease entity should be recognized. We report a case of surgically confirmed phlebosclerotic colitis in a 58-year-old woman with a history of long-term herbal ingestion.
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Key Words
Ischemic colitis;
Phlebosclerosis;
Calcification;
Mesenteric;
Herb

The term "phlebosclerotic colitis" was coined by Yao et al. in 2000.[1] Cases have rarely been reported, and most of the patients in these cases were Japanese, including the patient in the first case presented by Koyama et al. in 1991. The current literature suggests that the disease predominates in Asian populations. The symptoms of phlebosclerotic colitis are not specific and include abdominal pain, diarrhea, right flank pain, and tarry stools. Radiologic findings play an important role in the diagnosis of the disease. In most cases, patients eventually receive surgical intervention.

The etiology of phlebosclerotic colitis is still ill defined. Portal hypertension is thought to be a pathogenic factor,[1-5] but it does not account for some of the features of the disease.

Few cases have been reported in Taiwan.[5-9] Here, we report the case of a 58-year-old woman who was admitted to our emergency department with abdominal pain and right flank pain. Total colectomy and ileorectostomy were performed because of sepsis and peritoneal signs that were encountered after medical treatment during earlier hospitalizations.

Case Report

A 58-year-old woman was admitted to our emergency department in October 2009 with a 2-day history of right lower quadrant abdominal pain and right

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flank pain with frequent urination. She had received traditional herbal treatment, with regimens of 白求, 桔梗, 羌活, 柴胡, 甘草, 麻黃, 杏仁, 菜頭子, 石膏, 茯苓, 鈎陳, 黃芩, 沙參, 白芍, 生地 and 白芷, for sporadic diarrhea, headache, and upper respiratory tract infection for more than 20 years. She did not have a history of hepatitis, cirrhosis, renal colic, abdominal surgery, cardiovascular disease, or immunopathologic disorders. Her family history was unremarkable.

During the physical examination, the patient was afebrile and hemodynamically stable. Her vital signs were as follows: blood pressure, 117/76 mmHg; pulse rate, 80 beats/min; body temperature, 37.3 °C; and respiratory rate, 18 breaths/min. Neither pale conjunctiva nor icteric sclera was observed. The abdomen was flat and tender over the right lower quarter, without a palpable mass or a peritoneal sign. Right costovertebral angle knocking tenderness was noted.

Laboratory tests indicated a white blood cell count of 11,500 cells/μL, hemoglobin level of 13.2 g/dL, platelet count of 220,000 cells/μL, urinary white blood cell count of 80-85 cells/high power field (HPF), urinary red cell count of 2-3 cells/HPF, and fecal occult blood rating of 4 plus. Factors related to coagulation pathways, i.e., the prothrombin time, international normalized ratio, partial thromboplastin time, and protein C and S levels were all within normal limits. The D-dimer level was also unremarkable. Tumor markers, including α-fetoprotein, carcinoembryonic antigen, β-human chorionic gonadotropin, CA125, CA153, and CA199 were all within the normal range. The blood calcium (8.1 mg/dL; normal range: 8.6-10.2 mg/dL) and phosphorus levels (2.5 mg/dL; normal range: 2.7-4.5 mg/dL) were low. The levels of other electrolytes, i.e., glutamic oxaloacetic transaminase and glutamic pyruvic transaminase, were normal. No pathogens were detected in blood and urine cultures.

Multiple tortuous threadlike calcifications were observed along the course of the transverse colon and hepatic flexure on plain abdominal radiography (Fig. 1). Contrast-enhanced computed tomography (CT) revealed thickening of the wall of the ascending colon with increased density in the surrounding fatty tissue and calcification along the colonic and mesenteric vessels of the ascending colon and transverse colon (Fig. 1). Colonoscopy was not performed because the patient was not willing to undergo the procedure.

No surgical intervention was performed because the symptoms improved gradually with conservative treatment (enteric aspirin administration). After a 7-day hospital stay, the patient was discharged. After the first hospitalization, she was hospitalized again several times because of abdominal pain, fullness, and fever and was treated without surgery. In July 2011, total colectomy and ileorectostomy were performed because of extraluminal free air adjacent to the ascending colon observed by abdominal CT (Fig. 2) with peritoneal signs, bloodstream *Acinetobacter baumannii* infection-related sepsis, and unendurable abdominal pain. Multiple sclerotic plaques were found to be impacted in the colonic and mesenteric vessels

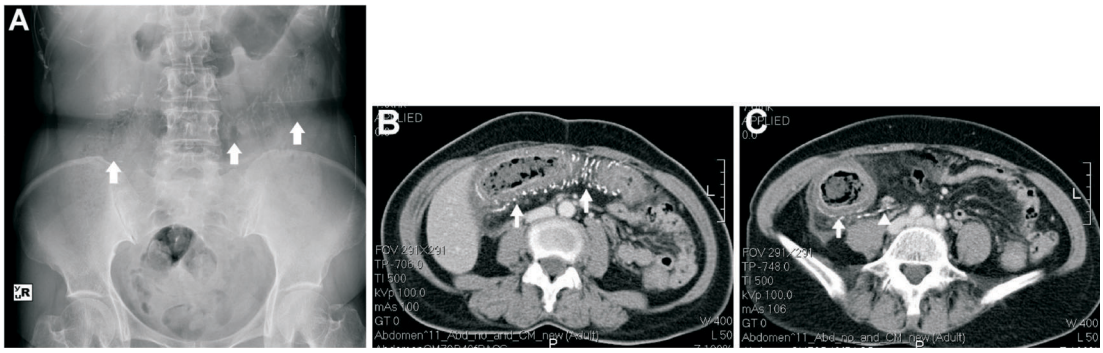

Fig. 1. (A) An abdominal roentgenogram shows multiple tortuous threadlike calcifications along the course of the transverse colon and hepatic flexure (arrows). (B) and (C) A contrast-enhanced computed tomography (CT) scan shows calcification along the colonic (arrows) and mesenteric (arrowhead) vessels over the ascending colon and the transverse colon with thickening of the colonic wall.

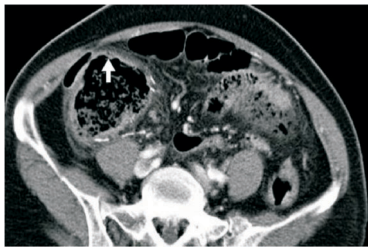

Fig. 2. A contrast-enhanced CT scan shows the extraluminal free air adjacent to the ascending colon (arrow).

of the involved colon (Fig. 3). Radiography of the resected colon showed calcification along the right colon and the descending colon. She was discharged 18 days after the surgery and has been in good health until now.

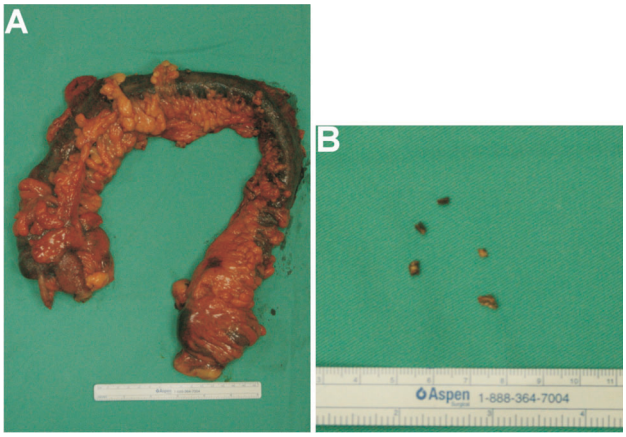

Fig. 3. (A) Dark purple coloration with a gangrenous appearance of the whole colon. (B) Multiple calcified materials removed from the vessels along the involved colon.

Discussion

Phlebosclerotic colitis is different from typical ischemic colitis because it is rarely encountered and always involves the right hemicolon (it occasionally involves the distal transverse colon and descending colon). Satio et al. found that 26 cases were reported in the English or Japanese literature until 2005.[10] Ten additional cases (including ours) have been reported in the English and Chinese literature thus far.[6-9,11-19] In most of these cases, the patients were from Japan, but some cases from Korea,[4] Hong Kong,[13] North America (the patient in this case was Taiwanese by birth),[14] and Taiwan have also been reported.

The first case was described by Koyama et al. in 1991. The term "phlebosclerotic colitis" was coined by Yao et al. in 2000,[1] and other authors have provided additional names such as "idiopathic mesenteric phlebosclerosis." Ying et al. presented the more precise term "chronic phlebosclerotic ischemic colitis," which indicates that phlebosclerosis is a characteristic, but not a cause, of the disease.[5]

Genetic factors may contribute to the disease because all of the patients in the cases reported thus far are of Asian descent. No concomitant disorders were shown to contribute to phlebosclerotic colitis, although portal hypertension was previously thought to be a possible cause.[1-5] The relationship between portal hypertension and phlebosclerotic colitis remains controversial. Markos et al. noted that phlebosclerotic colitis occurs independently of the calcification of the portal vein secondary to portal hypertension.[14] Among the reviewed cases, only a few showed evidence of cirrhosis, which gives rise to portal hypertension.[2,4] Ying et al. suggested that the poorly developed marginal arteries and collateral vascular network in the right hemicolon are the reason for the consistent involvement of this structure in the disease.[5] However, sufficient evidence to satisfactorily support a hypothesis regarding the pathology is lacking. In some studies, the cases were coincident with other disorders such as carcinoma and the CREST syndrome.[3,15] Chang summarized 5 cases including that of a couple with a history of herbal ingestion, which was similar to our case.[9] The hypothesis that herbal ingestion contributes to phlebosclerotic colitis requires further investigation.

The clinical course of phlebosclerotic colitis is chronic, and disease manifestation usually occurs months to years after the onset of symptoms because of the non-specific nature of symptoms such as recurrent diarrhea and abdominal pain as well as the lack of awareness of the clinicians. Nausea, vomiting, fever, right flank pain, tarry stool, constipation, and ileus often develop; however, these symptoms are not specific.

The finding for the conclusive diagnosis of the

disease has usually been calcification along the wall of the right hemicolon (the ascending and transverse colon) as seen on an abdominal roentgenogram or abdominal CT, except in some cases presented by Kusanagi et al. and Kang et al.[2,4] Calcification is typically observed but is not essential for the diagnosis of phlebosclerotic colitis, because it does not occur during the early stage of the disease.[2] In our case, the patient initially showed calcification along the right colon; the calcification was later found along the course of the descending colon in the pre-operation abdominal CT scan and post-operation radiogram of the resected specimen. Barium enema, superior mesenteric angiography, colonoscopy, histopathologic examination, virtual CT colonography, three-dimensional CT angiography,[13] magnetic resonance imaging, and magnetic resonance arteriography were also beneficial for the diagnosis or follow-up surveys.[13]

Most of the patients in the reported cases were treated surgically. The clinical symptoms subsided with conservative treatment in a few patients; however, surgeries were eventually performed for some patients because of serious relapses months or years later. Surgeries were not performed in only 16 of the 43 cases in the reviewed literature, but fewer patients may have actually achieved complete remission, because of insufficient elapsed time or lack of follow-up.[1,5-10,12,13,15-18] Even in previous studies with successful outcomes, the applied regimens were not reported to be absolutely adequate because the true etiology remains ill defined. In our opinion, surgery should be performed for patients with phlebosclerotic colitis because of the complications that are associated with medical treatment, such as peritonitis and sepsis.

The symptoms of phlebosclerotic colitis are not specific. The recognition of distinctive radiological features is critical for the diagnosis of phlebosclerotic colitis, which may otherwise be missed because of subclinical symptoms or the inexperience of clinicians. A colonoscopy, abdominal roentgenography, or abdominal CT should be performed for unexplained abdominal symptomatology. More cases need to be reviewed to determine the relationship between phlebosclerotic colitis and other diseases.

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病例報告

靜脈硬化性大腸炎：曾使用過中藥之病例報告

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靜脈硬化性大腸炎為缺血性大腸炎的一種，但不常見。其特徵是在右側大腸周圍會有鈣化形成。目前約有四十個病例被報導，大部分的病例皆接受手術。致病原因目前仍未清楚，雖然門靜脈高壓被認為是其中一個原因。臨床醫師應認識此疾病之特別的影像學特徵。我們報導一個手術證實為靜脈硬化性大腸炎且合併長期服用中藥過去史之五十八歲女性。

關鍵詞 缺血性腸炎、靜脈硬化、鈣化、腸繫膜、中藥。