Chronic Phlebosclerotic Ischemic Colitis

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Phlebosclerosis of the mesenteric vein is a rare cause of ischemic colitis. The clinicopathologic findings of chronic phlebosclerotic ischemic colitis are different from conventional ischemic colitis. We report a case showing the following features: (1) plain abdominal radiography-vascular calcifications at the ascending colon and transverse colon, (2) abdominal computed tomography (CT) - colonic wall thickening with intramural calcification and mesenteric venous calcification, (3) barium enema-luminal narrowing, wall rigidity and thumbprinting appearance, (4) superior mesenteric angiography-decreased vascularity of right colon, narrowing or occlusion of the marginal arteries, disturbance of venous return, dilatation and tortuosity of the veins along the vasa recta, (5) colonoscopy-dark purple-colored edematous mucosa and prominent submucosal veins, (6) colonoscopic biopsy-marked fibrous thickening with hyalinization of mucosal venous wall, narrowing of the lumen but no thrombus. The pathognomonic hallmark for radiological diagnosis of chronic phlebosclerotic ischemic colitis is the vascular calcifications in the colonic wall and mesenteric veins seen on plain abdominal radiography and abdominal CT.

Key words: Colitis, ischemic; Colon, CT; Veins, mesenteric; Veins, stenosis or obstruction

Ischemic colitis presents a wide clinical spectrum of severity. It may be caused by occlusion of main mesenteric arteries or veins, vasculitis, or decreased vascular perfusion without vascular lesions. Abnormalities of veins rarely cause ischemia of the bowel. Phlebosclerosis of the mesenteric vein is a rare cause of ischemic colitis. Few reports (only 19 cases) described this unusual chronic ischemic colonic lesion. We report here a case of chronic ischemic colonic lesion with phlebosclerosis and calcification of the mesenteric vein.

CASE REPORT

A 65-year-old female was admitted to our hospital in April 1998 because of right lower quadrant abdominal pain and abdominal bloating after meal for 6 months. Her medical history includes right nephrectomy at the age of 35, thyroidectomy at the age of 50 and cholecystectomy at the age of 61. Chronic viral hepatitis (type c) was diagnosed at 50 years of age.

Tenderness was noted in the right lower quadrant abdomen. No abnormalities were found in the complete blood count, urinalysis and stool examination. Elevation of alanine aminotransferase (41 IU/L, normal 35) and prolonged activated partial prothrombin time (28.3 secs, normal 27) were noted. The albumin was 3.4 g/dl and the globulin was 4.1 g/dl. There is no clinical or laboratory finding suggestive of portal hypertension. And there is no evidence of systemic collagen or vascular disease.

A plain abdominal radiography showed threadlike calcifications at the site of ascending colon and transverse colon (Fig. 1a). Some of the calcifications were arranged perpendicular to the bowel wall or parallel to the colon, which suggested vascular calcifications.

Abdominal computed tomography (CT) revealed wall thickening of the ascending colon and tranverse colon. Intramural calcifications and calcifications in the extracolonic branches of right and middle colic...
veins were noted (Fig. 1b,c). A barium enema study revealed poor distensibility, irregular contour, thumbprinting appearance and narrowing of the lumen of the ascending colon and transverse colon (Fig. 1d).

Superior mesenteric angiography showed decreased vascularity of right colon and narrowing or occlusion of the marginal arteries (Fig. 2a). In the early venous phase, disturbance of venous return and dilatation and tortuosity of the veins along the vasa recta in the ascending and transverse colon were noted. Tortuous and beaded appearance of the right colon mesenteric veins draining into the SMV main trunk was seen in the late venous phase (Fig. 2b). However, there was neither obstruction of the portal vein nor presence of collateral circulation.

Endoscopy of the upper gastrointestinal tract revealed no esophageal varices. Colonoscopy showed dark purple-colored edematous mucosa and prominent submucosal veins from the ascending colon to the transverse colon (Fig. 3a). An ulcer at the cecum was noted. Endoscopic biopsy specimen from the ulcer showed ulcer scar with granulation tissue formation, acute and chronic inflammation. Mucosal venous wall showed marked fibrous thickening with hyalinization and narrowing of the lumen but no thrombus (Fig. 3b). These features indicated chronic ischemic colonic lesion caused by phlebosclerosis with calcification.

Operation was not performed. Six months later, the colonoscopy follow-up showed only an ulcer at the transverse colon.

**DISCUSSION**

Phlebosclerosis is an adaptive change in the venous wall to prolonged and increased venous blood pressure, for example, owing to right-sided heart failure.
failure or portal hypertension [1,2]. Calcification of the portal vein and its tributaries is described in patients with portal hypertension and portal vein thrombosis [3,4]. There is no evidence of esophageal varices, splenomegaly and collateral circulation in our patient.

Intestine ischemia is caused mostly by abnormalities of arteries and venous thrombosis. Interruption to the colonic blood flow may be chronic or acute as a result of either a general state of hypoperfusion or local vascular obstruction [5]. There are 6 distinct clinico-pathological subtypes of ischemic colitis [6]. Our case is the subtype of chronic ulcerative ischemic colitis. Abnormalities of the veins rarely cause ischemia of the bowel. Churg-Strauss syndrome, Behcet's disease and lymphocytic phlebitis have been reported to cause mesenteric ischemia by selectively affecting the veins [7]. Phlebosclerosis of the mesenteric vein is a rare cause of ischemic colitis. It was first described by Koyama et al. in 1991 [8]. And the clinicopathologic findings of chronic ischemic colonic lesion caused by phlebosclerosis with calcification (chronic phlebosclerotic ischemic colitis) are different from those of conventional ischemic colitis [1,9].

Figure 2. a. SMA angiogram, arterial phase, shows decreased vascularity of the right colon and stenosis or occlusion of the marginal arteries (black arrows). b. In the early venous phase, disturbance of venous return of right colon is noted (not shown). Tortuous and beaded appearance of the right colon mesenteric veins draining into the SMV main trunk (white arrows) and no collateral circulation are seen in the late venous phase.

Figure 3. a. Colonoscopy of the ascending colon shows prominent submucosal veins (arrows). b. The cecal ulcer biopsy specimen shows marked fibrous thickening with hyalinization of mucosal venous wall (arrows).
affects the proximal colon slowly, initially, and produces chronic symptoms. Phlebosclerosis with calcification of the mesenteric veins is its dominant histopathological feature.

In the available literatures, only 19 cases were reported [1,7,9,10,11]. All cases were Japanese (7 men and 12 women; age range, 36-77 years). The clinical symptoms included abdominal pain, diarrhea, ileus, vomiting, constipation and bloody stool. The duration of illness was fairly long and the onset of symptoms was gradual. No common underlying diseases existed. Neither a common past medical history nor a common medication history was found. The right side colon was involved in all patients. In 1994, Ikehata et al. reported two cases who had gradual extension of disease to the distal colon over 5 years. And they concluded that chronic persistent ischemic colitis arises from result to phlebosclerosis as well as arteriosclerosis in the mesenteric system [7].

The characteristic findings of chronic ischemic colonic lesion caused by phlebosclerosis with calcification have been reported [1,7,9,11]. Our imaging findings are the same as those. The pathognomonic hallmark for clinical and radiological diagnosis of chronic phlebosclerotic ischemic colitis is the vascular calcifications in the colonic wall and mesenteric veins that revealed by plain abdominal radiography and abdominal CT [11]. Thumbprinting appearance of the affected colon seen on barium enema suggests intestine ischemia. The angiographic findings of chronic ulcerative ischemic colitis in our case are caused by phlebosclerosis as well as arterial occlusive disease of the colon. On the basis of clinical, radiologic, colonoscopic and histopathological findings of our patient, we could make the diagnosis of chronic phlebosclerotic ischemic colitis.

Colon cancer with calcification such as mucinous and scirrhous carcinoma has been reported [12,13], but the mesenteric venous calcification is not its image feature. In 1992, Yip et al. reported a case of calcification of arterial arcade of colon in a patient with ischemic colitis [14]. In our patient, it is not the arteriosclerotic calcification in the right and middle colic arteries. This can be proved from the CT and angiographic findings (Fig.1b,2a). Colonic calcification from Schistosomiasis may mimic chronic phlebosclerotic ischemic colitis. They are submucosal and subserosal calcified eggs and they mainly involve the inferior mesenteric vein territory [15,16], though they also can occur at right side colon such as in Schistosomiasis Japonicum [16].

Speculation concerning the etiology of chronic phlebosclerotic ischemic colitis includes idiopathic [11], watersoluble irritants, thrombotic phlebitis, mesenteric inflammatory venous-occlusive disease (MIVOD) [17], increased intraluminal pressure of the right-side colon [11], and CREST syndrome [18]. In the previous reported cases[ 9,11], authors paid too much attention to the venous abnormalities of phlebosclerosis with calcification and concluded that chronic ischemic colonic lesion was caused by phlebosclerosis. Unlike the left side colon, the right side colon frequently has poor-developed and inconsistent marginal arteries and collateral vascular network [19]. There are also different venous drainage pathways and collateral circulation between the right side colon and the left side colon [20]. That is why right side colon is involved in chronic phlebosclerotic ischemic colitis. We propose that chronic hypoxia (from arterial occlusive disease of the right side colon) and disturbed venous return and increased venous blood pressure (from chronic regional mesenteric inflammation due to chronic ulcerative ischemic colitis) may cause secondary phlebosclerosis with calcification of the mesenteric veins. So the more suitable nomenclature of this disease might be chronic phlebosclerotic ischemic colitis rather than chronic ischemic colonic lesion caused by phlebosclerosis with calcification.

Phlebosclerosis of the right side colon mesenteric vein is a rare cause of chronic ischemic colitis. The clinicopathologic findings of chronic phlebosclerotic ischemic colitis are different from those of conventional ischemic colitis. The pathogenesis remains unclear. The pathognomonic hallmark for radiological diagnosis of chronic phlebosclerotic ischemic colitis is the vascular calcifications in the colonic wall and mesenteric veins seen on plain abdominal radiography and abdominal CT.

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靜脈硬化合併鈣化所引起之慢性缺血性大腸炎：病例報告

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腸系膜靜脈硬化合併鈣化，是造成慢性缺血性大腸炎相當罕見的原因。我們報告一例因靜脈硬化合併鈣化所引起之慢性缺血性大腸炎。一位65歲女性，主訴右下腹疼痛，飯後腹脹，長達半年之久。其特有的影像及檢查特徵如下：(1) 腹部放射線攝影－在升結腸及橫結腸可以看出血管性之鈣化，(2) 腹部電腦斷層－大腸壁增厚及鈣化，並有腸系膜靜脈鈣化，(3) 下消化道攝影－大腸腔室變窄，腸壁變硬及指壓紋之腸壁外觀，(4) 上腸系膜動脈血管攝影－大腸之小動脈及周圍動脈狹窄或阻塞，靜脈回流受阻，(5) 大腸鏡－腸黏膜水腫，黏膜下腔靜脈擴張。

關鍵詞：缺血性大腸炎；大腸，電腦斷層；腸系膜血管；靜脈狹窄或堵塞