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## Single Case

# Possibility of Interstitial Lung Disease as a Phlebosclerotic Colitis Manifestation

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## Keywords

Phlebosclerotic colitis · Mesenteric phlebosclerosis · Interstitial lung disease

## Abstract

Phlebosclerotic colitis presents with ischemic bowels and calcification of the mesenteric veins. Owing to its rarity, we have little information on the complications of this disease. Herein, we report on a 77-year-old woman with phlebosclerotic colitis and interstitial lung disease. She was diagnosed as having phlebosclerotic colitis by CT and colonoscopy. At the same time, chest CT also showed interstitial lung disease. After 4 years, she experienced exacerbation of interstitial lung disease. She recovered without treatment. The occurrence of interstitial lung disease may have been associated with her phlebosclerotic colitis.

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## Introduction

Ischemic colitis is a common disease causing sudden abdominal pain and diarrhea in old patients, and characteristic of ischemia in the left side of the large intestine. Phlebosclerotic colitis also shows an ischemic pathology [1]. However, its clinical course and endoscopic findings are completely different from those of normal ischemic colitis. It is characteristic of transmural calcification of the colon [1] and is a rare disease, with the majority of reported cases in Asian populations [2]. Accordingly, there is a lack of data regarding complications of this disease. There is a possibility that interstitial lung disease is a rare manifestation of phlebosclerotic colitis. To our knowledge, this is the first case report on the possibility of such an association.

## Case Presentation

A 77-year-old woman developed gradual onset of abdominal pain without any symptoms of nausea, vomiting, constipation, or diarrhea. She presented to our hospital on the following day. A similar episode had occurred 1 year previously, with full recovery following a period of nil by mouth. She had a past history of phlebosclerotic colitis and interstitial lung disease, which had been diagnosed in our hospital 4 years previously. There was no remarkable family history. She had been taking herbal medicine, including gardenia fruits, *Coptis japonica*, *Scutellaria* root, and *Phellodendron* bark, for 4 years before the first episode of phlebosclerotic colitis.

All her vital signs were normal. Chest examination revealed no rales. She had lower abdominal pain but no evidence of guarding or rebound tenderness.

Her white blood cell count was  $12.6 \times 10^9/L$ . There were no other abnormal findings on complete blood cell count or blood chemistry analysis. Chest plain radiography revealed reticular shadowing in both lower lung fields (Fig. 1a), and chest CT demonstrated scattered reticular shadows (Fig. 1b). Abdominal plain radiography revealed linear calcification in the ascending colon (Fig. 2a). Abdominal CT demonstrated thickening of the ascending colon and calcification of the mesenteric vessels. The small intestine was seen to be dilated. No findings suggesting portal hypertension were observed (Fig. 2b). Colonoscopy showed dark purple edematous sclerotic mucosa from the transverse colon to the cecum (Fig. 3). A microscopic examination of the ascending colon revealed proliferation of collagen fibers. We observed moderate hyalinization adjacent to capillaries (Fig. 4). Phlebosclerotic colitis was diagnosed according to the results of pathological examination.

The patient was admitted to our hospital, and herbal medicine was discontinued. She recovered with conservative management. She was discharged 1 week after admission.

Two months later, she developed gradual onset of dyspnea. Chest CT demonstrated thickening of the interlobular septa and panlobular ground-glass opacities in both lungs (Fig. 1c). Abdominal CT again revealed bowel wall thickening from the ascending to the transverse colon; however, the small intestine was no longer seen to be dilated (Fig. 2c). Two months later, her dyspnea gradually deteriorated and bronchoscopy was planned. On admission, repeat chest CT was performed, demonstrating resolution of the previously observed pulmonary shadowing (Fig. 1d). Her dyspnea also gradually ameliorated without treatment. Microbial analysis of bronchoalveolar lavage fluid resulted negative. Cytologic analysis of bronchoalveolar lavage fluid revealed a composition of 44% lymphocytes, 7% eosinophils, 2% neutrophils, and 47% other cell types.

The patient was discharged on the day following bronchoscopy. No recurrence of phlebosclerotic colitis or interstitial lung disease was observed after 4 months of follow-up.

## Discussion

The etiology and pathogenesis of phlebosclerotic colitis are not clearly understood, but they may be associated with thrombosis, atherosclerosis, portal hypertension, or immunological abnormality [3]. Phlebosclerotic colitis has been shown to be caused by a decreased blood flow to the mesenteric veins [4]. This disturbance of venous return may also occur in other organs.

Using PubMed, we searched for case reports using the keywords “phlebosclerotic colitis” and “mesenteric phlebosclerosis” on July 18, 2016. We identified 28 articles and 41 cases reported between 1997 and 2015; however, none were reported to have interstitial lung disease. Nakayama et al. [4] reviewed 37 reports written in Japanese from 1983 to 2009 and identified no cases with interstitial lung disease. We also searched for case reports written in Japanese on July 18, 2016. By searching Ichushi, a Japanese medical database, using the keywords “phlebosclerotic colitis” and “mesenteric phlebosclerosis,” we identified 28 articles and 34 cases reported between 2010 and 2015; however, none of these cases were reported to have interstitial lung disease either.

Chest CT typically reveals butterfly shadowing with venous return disturbance in cases of interstitial lung disease. However, we speculate that interstitial lung disease may have been associated with phlebosclerotic colitis in the present case. First, there were no other factors present that may have caused secondary interstitial pneumonia. Both phlebosclerotic colitis and interstitial lung disease could be side effects of the herbal medicine. Gardenia fruits are particularly reported to be related to phlebosclerotic colitis [5]. However, the patient had discontinued taking herbal medicine 2 months before the exacerbation; therefore, it is unlikely that the exacerbation of interstitial lung disease was caused by the herbal medicine. Second, in the present case, interstitial lung disease followed a medical course similar to that before phlebosclerotic colitis and it resolved without treatment. Third, chest CT demonstrated thickening of the interlobular septa and panlobular ground-glass opacities, findings consistent with decreased blood flow to the pulmonary veins, as occurs in the mesenteric veins in cases of phlebosclerotic colitis [5]. Panlobular ground-glass opacities are mainly found with sinopulmonary diseases such as pneumonia, but they are also observed with pulmonary edema caused by decreased blood flow.

In the present case, pulmonary shadowing appeared 2 months after resolution of the abdominal pain. Furthermore, transbronchial lung biopsy was not performed. Further follow-up is required to determine whether phlebosclerotic colitis and interstitial lung disease follow a similar medical course in the present case.

To our knowledge, this is the first report of phlebosclerotic colitis complicated by interstitial lung disease. Interstitial lung disease may be a rare manifestation of phlebosclerotic colitis. Clinicians should be aware of interstitial pneumonia in patients with phlebosclerotic colitis.

## Statement of Ethics

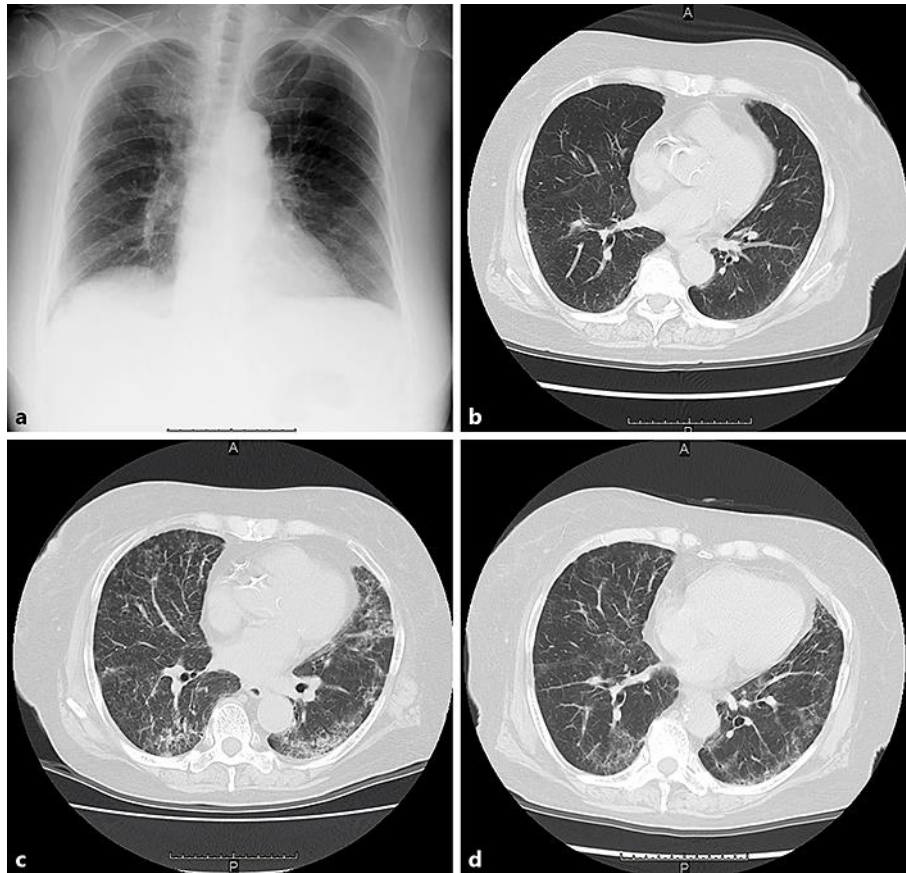
The authors have no ethical conflicts to disclose.

### Disclosure Statement

The authors have no conflicts of interest to disclose.

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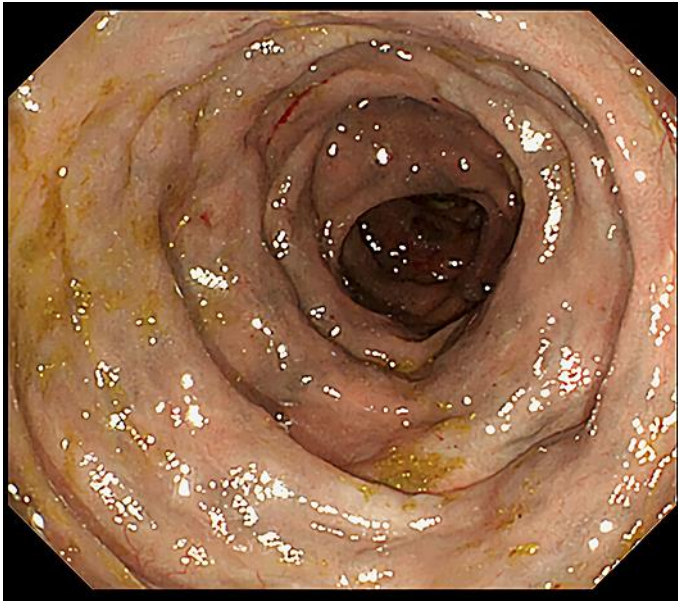
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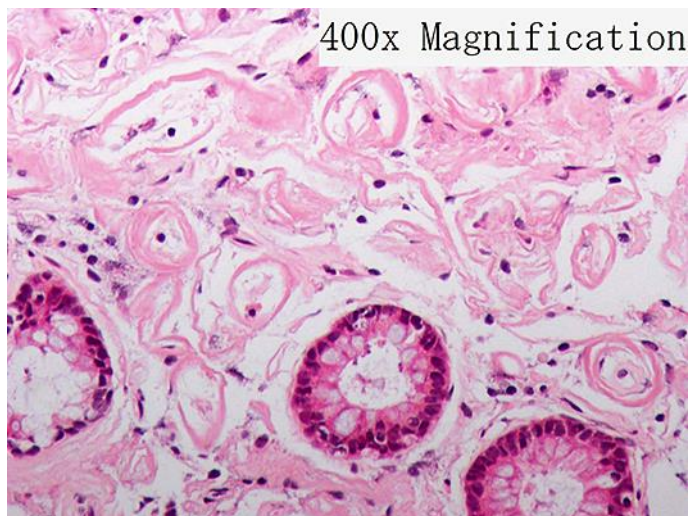
**Fig. 1.** Timeline of chest CT and plain radiography findings. **a** Plain chest radiograph at the time of first admission showing reticular shadowing of both lower lung fields bilaterally. **b** High-resolution CT (HRCT) image of the chest at the time of first admission showing scattered reticular shadows. **c** HRCT image of the chest 2 months later showing exacerbation of the ground-glass opacities and thickening of the interlobular septa. **d** HRCT of the chest at the time of the second admission showing resolution of the ground-glass opacities.



**Fig. 2.** Timeline of abdominal CT and plain radiography findings. **a** Plain abdominal radiograph at the time of first admission showing linear calcification in the ascending colon (arrowheads). **b** CT of the abdomen at the time of first admission showing thickening of the ascending colon and calcification of the mesenteric vessels (arrowheads). The small intestines are seen to be dilated (arrows). **c** CT of the abdomen 2 months later showing thickening of the bowel walls from the ascending to the transverse colon and calcification of the mesenteric veins (arrowheads). The small intestine no longer appears dilated.



**Fig. 3.** Representative colonoscopy image at the time of first admission demonstrating the presence of dark purple edematous mucosa; this was seen to extend from the transverse colon to the cecum.



**Fig. 4.** Representative microscopic examination image at the time of first admission demonstrating proliferation of collagen fibers and moderate hyalinization adjacent to capillaries.