INTERNAL MEDICINE

□ CASE REPORT □

Colonic Sarcoidosis Presenting Multiple Submucosal Tumor-Like Lesions

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Abstract

Here, we describe a case of colonic sarcoidosis that developed over a 7-year period of observation of intrathoracic sarcoidosis. The patient was asymptomatic, but colonoscopy showed multiple elevated lesions mimicking submucosal tumors in several areas of the colon. The specimens obtained by biopsy showed noncaseating granuloma, suggesting sarcoidosis. The observations in the present case indicate that colonic involvement should be considered in patients with sarcoidosis. Furthermore, the macroscopic appearance of multiple submucosal tumor-like lesions in colonic sarcoidosis is extremely rare.

Key words: colon, sarcoidosis, submucosal tumor

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Introduction

Sarcoidosis is a systemic granulomatous disease that primarily affects the lungs and lymphatic systems (1). The disease also involves the eyes, skin, liver, and spleen (2, 3). The incidence of gastrointestinal (GI) tract involvement is less than 1.0% (1, 4). However, some studies have shown evidence of subclinical GI tract sarcoidosis in 5-10% patients with systemic sarcoidosis (2, 5). The stomach is the most commonly involved part of the GI tract and the colon is involved less frequently (1, 6). In the literature, colonic sarcoidosis has been reported to show several nonspecific endoscopic findings, including plaque-like lesions, ulcers, friable mucosa, fold thickening, focal nodularity, segmental narrowing, or normal mucosa (6). Here, we report a case of colon involvement that developed during the period of long-term observation of pulmonary sarcoidosis. The colonoscopic findings showed multiple elevated lesions mimicking submucosal tumors in several areas of the colon. These colonoscopic findings are extremely rare. Here, we present a case report and a review of the relevant literature.

Case Report

In July 2007, a 64-year-old woman with a 7-year history of known sarcoidosis visited our hospital because of blood in the stool detected on routine health screening. She had no specific abdominal or thoracic symptoms. At the initial diagnosis of sarcoidosis, chest radiograph showed bilateral hilar lymphadenopathy (Fig. 1a) and chest computed tomography (CT) showed slight interstitial shadow in the bilateral lower lobe (Fig. 1b). Laboratory examination showed an elevated level of angiotensin-converting enzyme (32.7 U/mL; normal, <25.0). The diagnosis of sarcoidosis was made based on the histological findings obtained from a transbronchial lung biopsy specimen (Fig. 1c). The patient was observed without medication. Physical examination, including ophthalmological examination, revealed no significant findings. Chest radiograph showed lymphadenopathy which was relatively similar to that in the initial diagnosis (Fig. 2a), but chest CT revealed that the pulmonary fibrosis had progressed (Fig. 2b). Total colonoscopy revealed multiple submucosal lesions with a tumor-like appearance in the ascending colon and transverse colon. The lesions were sessile, soft, smooth surface and measuring 2-5 mm in diameter (Fig. 3a, b). En-

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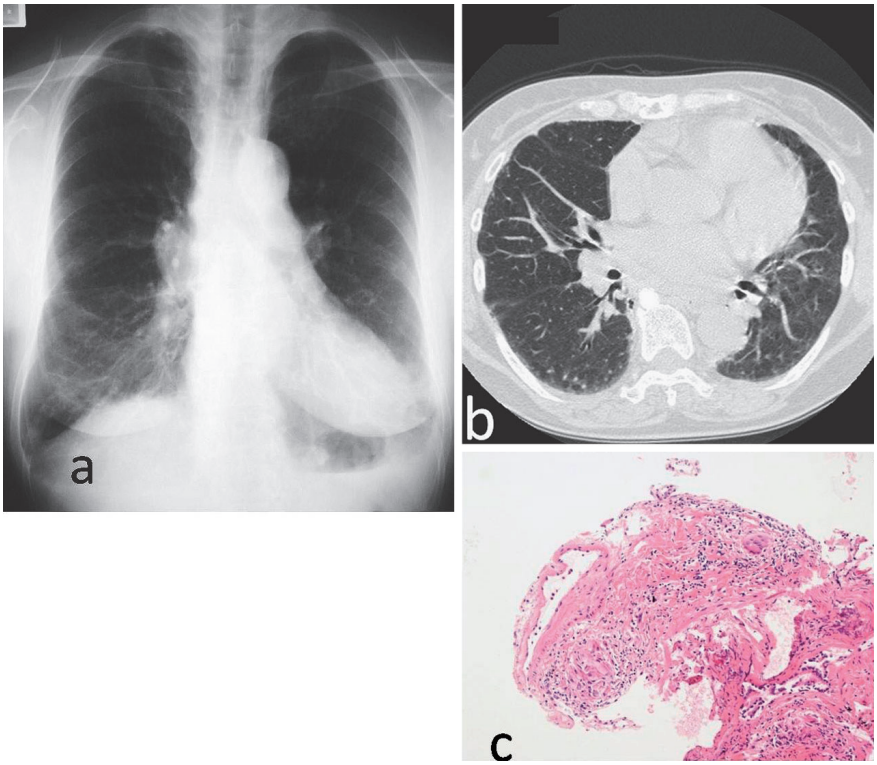

Figure 1. **(a) Chest radiograph at the initial diagnosis showed bilateral hilar lymphadenopathy. (b) Chest computed tomography (CT) at the initial diagnosis showed pulmonary fibrosis in the bilateral lower lobe. (c) Histopathology of the lung specimens obtained by the transbronchial lung biopsy showed non-caseating epithelioid cell granulomas with Langhans giant cells.The biopsy specimen showed multiple granulomas (original magnification, ×200, Hematoxylin and Eosin staining).**

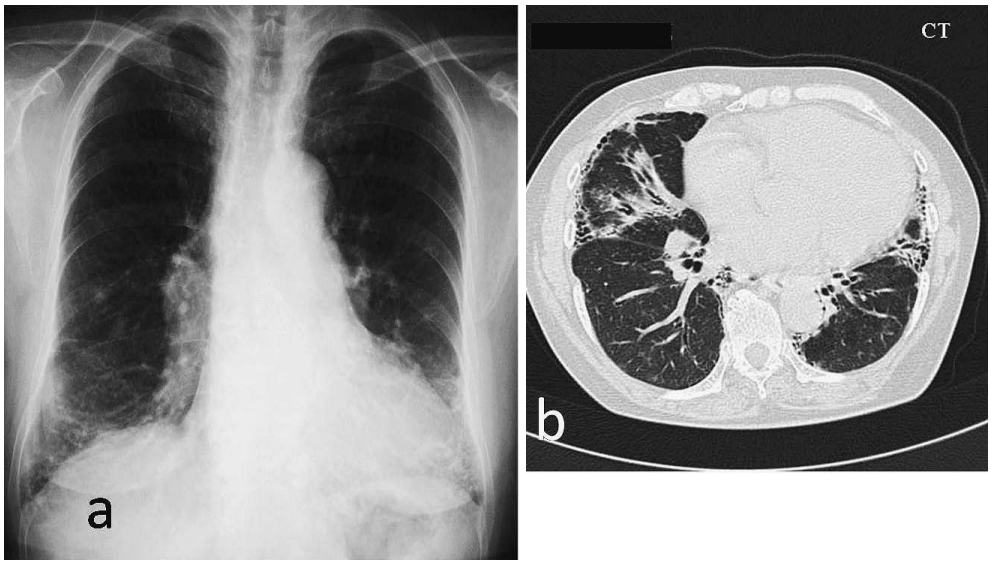

Figure 2. **(a) Chest radiograph seven years after the initial diagnosis revealed lymphadenopathy as shown in the initial diagnosis. (b) Chest CT seven years after initial diagnosis showed that the slightly advanced pulmonary fibrosis had progressed.**

doscopic ultrasound findings of the lesions in the ascending colon showed the hypoechoic tumors under the muscularis mucosae (Fig. 3c). Endoscopic resection was performed and the specimens showed multiple noncaseating granulomas, suggesting sarcoidosis (Fig. 4a, b). There were no signs of other colonic granulomatous disorders, such as tuberculosis, syphilis, fungal infection, or Crohn's disease. Examination of bronchoalveolar lavage fluid indicated an elevated lym-

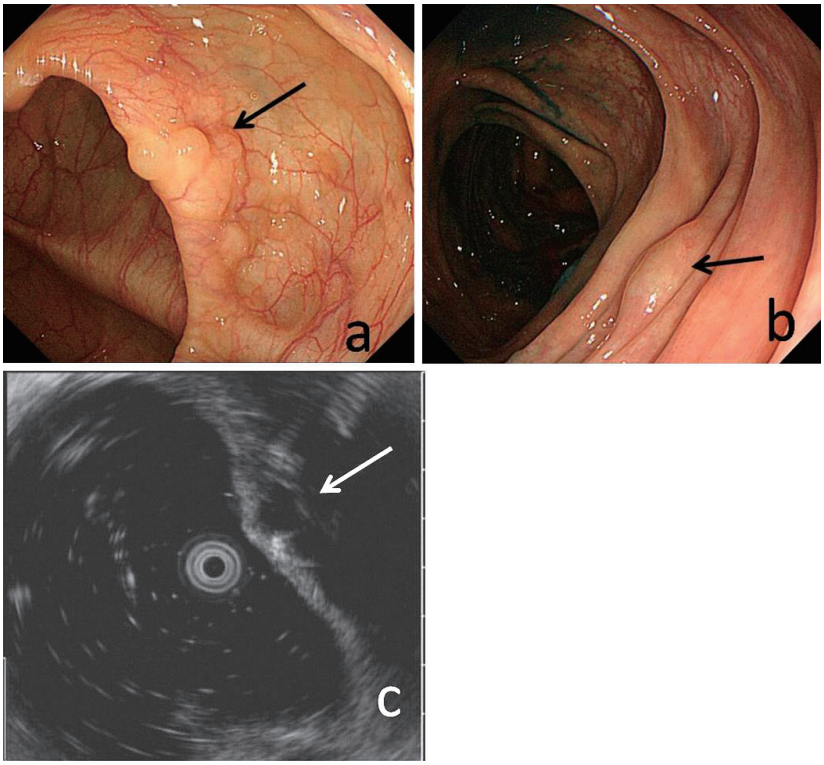

Figure 3. **Colonoscopy findings showed multiple submucosal tumor-like lesions in the ascending colon (a) and transverse colon (b) (black arrows). The lesions were sessile, soft, smooth surface and measuring 2-5 mm in diameter. (c) Endoscopic ultrasound findings of the lesions in the ascending colon showed the homogenous hypoechoic mass under the muscularis mucosae (white arrow).**

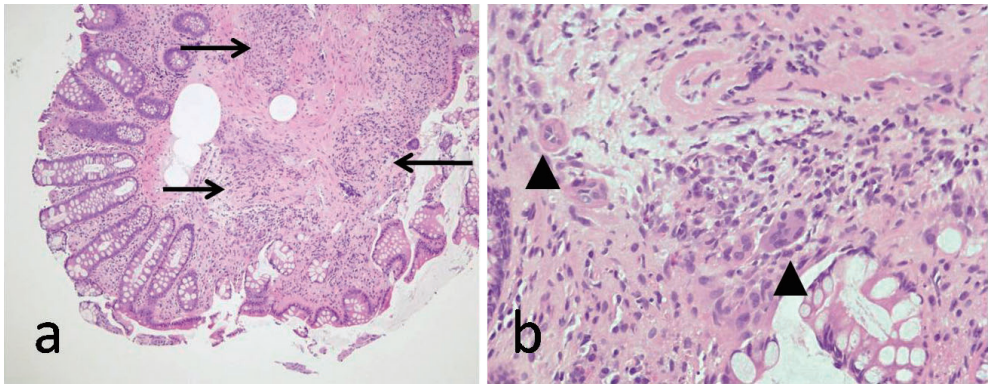

Figure 4. **Hematoxylin and Eosin staining of the submucosal lesions with a tumor-like appearance in the ascending colon biopsy specimens at low magnification (× 100) (a), and at high magnification (× 400) (b). Noncaseating epithelioid cell granuloma (arrows) and multinuclear Langhans giant cells (arrowheads) was seen.**

phocyte count (37%) and the ratio of CD4/8 lymphocytes was high (4.01). Chest radiography and computed tomography showed bilateral hilar lymphadenopathy, suggesting progression compared to the initial diagnosis of sarcoidosis.

Discussion

The actual incidence of GI tract involvement of sarcoidosis is unknown, but this association rarely appears to be clinically significant. The incidence of GI tract involvement is less than 1.0% (1). The stomach is the most commonly involved part of the GI tract, followed by the colon (7).

Nchimi et al. (7) reported a case with thickening of the terminal ileum wall, which caused intermittent diarrhea. Aaronson et al. (8) reported a case of sigmoid colonic sarcoidosis presenting with constipation and hematochezia. The lesion was localized stenosis with granulomatous inflammation of the mucosa, submucosa, and diffuse submucosal fi-

brosis. In addition, Hilzenrat et al. (9) reported a case with stricture and obstruction of the colon, resulting in abdominal pain, distention, vomiting, constipation, and weight loss. In these cases, clinical symptoms and colonoscopy findings were present. Furthermore, Veitch and Badger (10) reported asymptomatic colonic sarcoidosis presenting as colonic polyposis. These polyps had the appearance of adenomatous polyps.

The present case showed multiple elevated lesions mimicking submucosal tumors in the colon. This case represents a highly unusual presentation of sarcoidosis with colonic involvement. As described in other reviews of colonic sarcoidosis, radiographic and endoscopic findings provided no specific diagnostic information to distinguish this from other colonic diseases. The macroscopic appearance in the present case also did not differ from those of other types of colonic submucosal tumor. We found histological evidence of sarcoid granules in these lesions. As we failed to find sarcoid granules from apparently normal mucosa, we speculate that the colonic sarcoidosis first involved the submucosal lymph tissue, and the lesion then spread and grew to a submucosal tumor. Thus, the diagnosis of colonic sarcoidosis is dependent on the histological evidence of sarcoid granuloma. On the other hand, several cases of colonic sarcoidosis presenting with normal mucosa have been reported. The colonic involvement of sarcoidosis was found incidentally in grossly normal mucosa (6). Accordingly, we emphasize that histological examination is necessary to establish or exclude a diagnosis of colonic involvement in patients with sarcoidosis.

The colonic involvement in the present case appeared to parallel the progression of the thoracic lesions, suggesting that the colonic disease reflected advanced systemic sarcoidosis. However, Kaneki et al (11) reported that gastric sarcoidosis became apparent after the disappearance of ocular and intrathoracic disorders. Thus, each organ involvement in patients with sarcoidosis is very complex during the clinical course. Unfortunately, we had no endoscopic findings or evidence of histological involvement of the colon in our case before the present diagnosis. Therefore, it is necessary to evaluate thoracic and colonic lesions in future cases.

In summary, we presented a case of colonic sarcoidosis with multiple submucosal tumor-like lesions. As colonic sarcoidosis is a rare clinical manifestation, it is necessary to consider the possibly of colon involvement in patients with sarcoidosis.

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