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A Diagnostic Conundrum in a Newly Diagnosed Ulcerative Colitis Patient Who Presented with Pleuropericardial Effusion

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Abstract

Ulcerative colitis is a chronic inflammatory disease affecting mainly the colon and presenting with diarrhea, bloody defecation and abdominal pain. Although cardiac and/or pulmonary involvement has been reported in patients with ulcerative colitis, it rarely involves both the pleura and pericardium at the same time. Also, it is difficult to determine whether pulmonary or cardiac disease is secondary to the ulcerative colitis drugs or to the underlying disease process. Here we present a rare case of pleuropericardial effusion in a patient newly diagnosed with ulcerative colitis. In ulcerative colitis, the simultaneous involvement of the respiratory and cardiovascular systems is uncommon yet potentially dangerous.

Keywords: Pleuropericardial Effusion, Ulcerative Colitis, Mesalazine

1. Introduction

Ulcerative colitis (UC) is a chronic inflammatory disease commonly presenting with diarrhea, bloody defecation and abdominal pain. Extraintestinal manifestations of UC can involve the skin, eyes, joints, biliary tract, nervous system, heart and lungs (1-4). Although cardiac and/or pulmonary involvement has been reported in patients with UC, the disease rarely involves both the pleura and pericardium at the same time (4-6). Understanding the pattern of UC-associated extraintestinal manifestations, such as its pulmonary and cardiac effects, is critical for securing optimal management (1). Given that presentation is uncommon and variable, UC-related respiratory and cardiac manifestations are often easily overlooked (3, 6). These involvements are generally not directly related to the intestinal activity of the disease (7); however, the rate of occurrence of extraintestinal manifestation increases with duration of the intestinal disease (8). Pulmonary involvement was first reported in 1976, with many cases having been diagnosed to date (9). Among UC patients, cardiac involvement has rarely been reported. In the UC population, pericardial effusion and pericarditis are the most frequently reported extraintestinal cardiac manifestations (10). The pathophysiology of UC-associated pulmonary and cardiac disease remains obscure (3, 8). The same cardiac and pulmonary complications are related to UC treatment, especially with sulfasalazine and mesalazine (5, 10-14). Nevertheless, it is still difficult to determine whether pulmonary or cardiac disease is secondary to the UC drugs or to the underlying disease process. Herein we present a rare case of pleuropericardial effusion in a patient newly diagnosed with UC.

2. Case Presentation

A 24-year-old Caucasian man was hospitalized because of persistent bloody diarrhea, weight loss and abdominal pain. During colonoscopic examination, he was diagnosed with left sided Grade II UC (Mayo Clinical Scoring). Colonic biopsy specimen reports confirmed the diagnosis, and the patients was commenced on 3 g/day of mesalazine. After improvement in symptoms, the patient was discharged to be followed up as an outpatient. On the twentieth day of therapy, the patient was re-hospitalized due to fatigue, fever and bloody defecation. He was febrile (40°C) and had tachycardia. Arterial blood pressure was 100/60 mmHg and pulse rate was 108 beats/min. He had normal heart and lung sounds, but hyperactive bowel sounds. Abdominal tenderness and rebound tenderness were not present.

Levels of liver enzymes, urea-creatinine, and serum electrolytes were within the normal ranges. However, albumin level was 1.90 g/dL (3.5 - 5.0), potassium level was 2.8 mmol/L (3.5 - 5.3), white blood cell count was 23.8 uL (4.0 - 10.5), C-reactive protein (CRP) was 139 mg dL (0 - 8), and erythrocyte sedimentation rate was 102 mm/h (0 - 15). Thoracoabdominal computerized tomography findings were normal. Again, Grade II UC (Mayo Clinical Scoring) was observed via colonoscopy. Stool, urine and blood culture samples were obtained. On the fifth day, shortness of breath and chest pain were also added to the patient's ongoing complaints. On physical examination, decreased sounds were found in the basal area of the left lung, and a chest X-ray revealed the blunting of the left costophrenic angle, consistent with left pleural effusion (Figure 1A). Mild to moderate pericardial effusion was detected via echocardiography (Figure 1B). After performing thoracentesis, exudative fluid was diagnosed; pleural samples and sputum were cultured, but came back negative along with all other cultures. Histopathological examination of pleural samples was also negative. The autoimmune profile including anti-nuclear antibody, anti-double stranded DNA and anti-nuclear cytoplasmic antibodies was negative. The patient's clinical and laboratory investigations had no evidence of infection, tuberculosis, autoimmune disease or malignancy. Finally, we thought that the pleuropericardial effusion was related to UC and/or mesalazine. Mesalazine was ceased, and diclofenac sodium (50 mg tablet; BID) was commenced. Three days later, the patient became afebrile and the complaints were relieved; complete recovery (symptomatic and radiological) was achieved ten days after changing treatment regimen (Figure 1C). We decided to rechallenge with mesalazine treatment, and restarted mesalazine tablets at a dose of 1.5 g/day. At first presentation, the symptoms might have been precipitated by either the drug or UC itself. After the rechallenge, the findings were classified as an extra-intestinal manifestation of UC due to the absence of any other causative factors. No chest symptoms recurred, and the patient was discharged with 3 g/day of mesalazine a few days later. To date, the patient has been followed as an outpatient treated with the same dose of mesalazine.

3. Discussion

Although extraintestinal manifestations of UC are relatively common, clinically obvious pulmonary or cardiac disease is rare. Moreover, concomitant pleural and pericardial effusions are very rare conditions in patients with UC (3). UC-associated pulmonary disease can present as an airway (from the trachea to the bronchioles), lung parenchymal, or pleural disease. Pulmonary function test abnormalities may occur and, less commonly, the pulmonary vasculature may be involved (15). Pleural effusion can be directly or indirectly related to UC, occurring as a disease complication or being triggered by a related infection or pharmacological therapy (3). Most UC patients with pleural effusion are young males; this condition can occur during either the inactive or active phase of the disease (15). UC-related pleural manifestations can be classified as pleural effusion, pleuritis, pneumothorax, and pleural thickening (3, 16). Pleural effusion directly related to UC is usually unilateral, exudative, and may be hemorrhagic (15). It is critical to evaluate pleural effusion and rule out other etiologies before making an exact decision. UC-related cardiac extraintestinal manifestations have been less commonly reported. Pericarditis and pericardial effusion represent as frequent cardiovascular manifestations (70% of the total number of cardiovascular complications) (17-19). The prevalence of pericarditis is 0.19% among Crohn's disease patients and 0.23% among UC patients (17). A review of 68 patients with IBD revealed that pericarditis occurs more frequently in male patients with UC (18). These cardiac involvements include pericardial effusion, cardiac tamponade, acute pericarditis, myopericarditis, myocarditis, conduction delays and heart block (20, 21). Pericardial effusion and acute pericarditis are the more commonly seen cardiac complications, and patients with extended colonic inflammation have a higher risk of pericardial involvement. Pericardial involvement generally happens with disease flares. This cardiac manifestation may also take place independently of UC activity. Transesophageal echocardiogram plays a pivotal role in the diagnosis of UC-associated cardiac involvement. Pericardial complications can either be directly related to UC or to its treatment (20, 21).

UC patients with pleuropericardial effusion should be examined for malignant, infectious, and autoimmune agents as well as assessing their medications. If no findings of such etiologies exist, then pleuropericardial effusion can be defined as an extra-intestinal manifestation of UC. Previously, it has been shown that mesalazine itself can also cause pleural and/or pericardial effusion (5). Actually, it is not easy to discriminate mesalazine-related pleuropericardial effusion from UC-related pleuropericardial effusion because their clinical findings are very similar. Mechanisms recognized as causes of mesalazine-induced pleuropericardial effusion can be categorized as plasma concentration dependent and independent mechanisms (5, 13). On the other hand, this rare side effect can be related to immune-mediated mechanisms (13). Moreover, it has been reported that desensitization can be achieved with low-dose mesalazine treatment (5). Initially, we believed that mesalazine might have caused the pleuropericardial effusion in our case, so we ceased mesalazine treat-

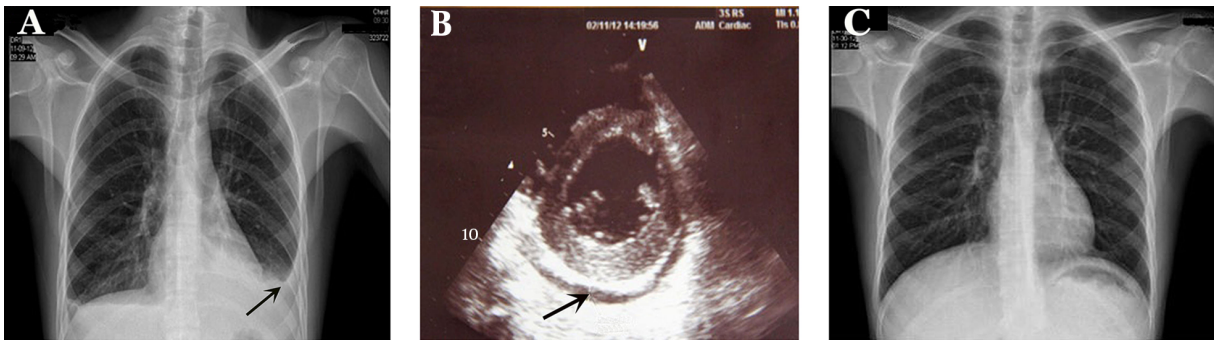

Figure 1. A, pleural effusion visible on chest X-ray; B, diagnosis of pericardial effusion by echocardiography; C, radiological improvement of pleural effusion

ment and began diclofenac sodium. After achieving clinical improvement, a low-dose mesalazine rechallenge was performed; no chest symptoms were reported and the patient was continued on high dose mesalazine tablets. However, as we mentioned above, it is not easy to discriminate mesalazine-related pleuritis from UC related-pleuritis. This rare condition is generally responsive to anti-inflammatory medications such as steroids and NSAIDs (5, 6). Also, in our case, the favorable results after commencing NSAID therapy confirmed this idea.

Pulmonary and cardiac manifestations of UC are becoming more and more recognized. The involvements of the respiratory and cardiovascular system in UC are uncommon yet can be potentially dangerous. In the case of dyspnea and other chest symptoms, patients with UC, especially those on mesalazine treatment, should be suspected for pleuropericardial effusion. Cessation of mesalazine and replacement with steroids or NSAIDs is recommended, after which patients should be closely followed and low-dose mesalazine rechallenge can be performed.

Footnotes

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