Case Report

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Acute Respiratory Distress Syndrome With Alveolar Hemorrhage due to *Strongyloidiasis* Hyperinfection in an Older Patient

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Strongyloides stercoralis is an intestinal nematode that occurs sporadically in temperate areas like Korea. People who are in the immunosuppressed state, over the age of 65 or under the corticosteroid therapy are at risk for developing *Strongyloides* hyperinfection syndrome. Acute respiratory distress syndrome (ARDS) with alveolar hemorrhage is a rare presentation of *Strongyloides* hyperinfection. A 78-year-old man had been irregularly injected corticosteroid on his knees, but did not have any immunosuppressive disease. He was initially diagnosed with ARDS and septic shock. Bronchoalveolar lavage (BAL) fluid was bloody and its cytology revealed helminthic larvae identified as *S. stercoralis*. Results of *Cytomegalovirus* polymerase chain reaction (PCR), *Pneumocystis jirovecii* PCR, and *Aspergillus* antigen testing of the BAL fluid were positive. The clinical progress quickly deteriorated with multiple organ failure, shock and arrhythmia, so he finally died. This is a rare case of ARDS in an older patient without any known immunosuppressive conditions, with alveolar hemorrhage and *S. stercoralis* being found via BAL. **(Ann Geriatr Med Res 2018;22:200-203)**

Key Words: *Strongyloides stercoralis*, Acute respiratory distress syndrome, Alveolar hemorrhage

INTRODUCTION

Strongyloides stercoralis is an intestinal nematode that causes an endemic disease in tropical and subtropical rural areas.[1] In Korea, which is in a temperate zone, several cases of *Strongyloides* hyperinfection have been reported, but these have been mostly associated with gastrointestinal involvement.[2-4] Two cases of pulmonary strongyloidiasis with acute respiratory failure and alveolar hemorrhage[5,6] have been reported, but these cases involved an immunosuppressed state owing to lung cancer and chemotherapy. I report a case of acute respiratory distress syndrome (ARDS) in an older patient without known immunosuppressive conditions, with alveolar hemorrhage and *S. stercoralis* being found via bronchoalveolar lavage (BAL).

CASE REPORT

A 78-year-old man visited the Daegu Catholic University Medical Center owing to aggravated dyspnea. He had diarrhea 10 days before admission, and dyspnea started 3 days before admission. He was previously diagnosed with hypertension and degenerative arthritis and intermittently received intra-articular injections of corticosteroids into his knee joints. He was a farmer and had never traveled overseas.

His arterial blood pressure was 110/60 mmHg, pulse rate was 103 beats/min, body temperature was 38.9°C, and respiratory rate was 28/min. Crackling was heard in both lower lung fields. A complete blood count showed a leukocyte count of 8,300/μL (90.4% neutrophils, 6.1% lymphocytes, and 0.7% eosinophils), a hemoglobin level of 9 g/dL, and a platelet count of 200×10^3/μL. Laboratory tests showed the following blood levels: aspartate aminotransferase, 66 U/L; alanine transaminase, 137 U/L; gamma-glutamyltransferase, 90 U/L; blood urea nitrogen, 23.6 mg/dL; creatinine, 0.6 mg/dL; Na, 131 mEq/L; K, 3.7 mEq/L; total protein, 4.8 g/dL; and albumin, 2.6 g/dL. The levels of pro-B-type natriuretic peptide and C-reactive protein at 1,414 pg/mL (normal range, 9–113.2 pg/mL) and 368 mg/L (normal range, <5 mg/L), were increased. The result of anti-HIV antibody screening was negative. The result of stool occult blood testing was positive, but no parasites or eggs were found in the stool. Arterial blood gas analysis showed pH 7.500, pCO_2 22.5 mmHg, pO_2 84.4 mmHg, HCO_3 17.2 mmol/L, and peripheral oxygen saturation (SpO_2) 97.3% under a fraction of inspired oxygen (FiO_2) of 0.6.

No microorganisms were detected in blood and sputum cultures. Results of a sputum acid fast bacillus (AFB) smear and *Mycobacterium tuberculosis* polymerase chain reaction (PCR) were negative.

Chest radiography revealed bilateral and diffuse nodular infiltration and multiple patchy opacities in both whole lung fields and a cavity in the left hilar area (Fig. 1A). Chest computed tomography (CT) showed multiple consolidation, ground-glass opacity, and centrilobular nodules in both lungs and several thin-walled cavities in the right upper lobe and left lower lobe, with septa inside the cavities (Fig. 1B). Abdominal CT showed diffuse bowel wall edema along the ascending and transverse colon.

As a result of the above findings, ARDS, pneumonia, and sepsis were diagnosed, and treatment with piperacillin/tazobactam and ciprofloxacin was initiated along with oxygen therapy. At 14 hours after admission, as the patient's SpO_2 decreased to 83% and his respiratory rate increased to 32 breaths/min, intubation and ventilator treatment was initiated. Hemoptysis was confirmed from the time of tracheal intubation and persisted thereafter. On the first day of admission, the patient's blood pressure had decreased, and he was treated with fluid and inotropic support based on the diagnosis of septic shock. On the third day of admission, bronchoscopy was performed and showed large blood clots in both bronchi and diffuse alveolar hemorrhage in the lower parts of the bronchi when the blood clot was removed (Fig. 2A). BAL was performed from the right upper lobe bronchus, where cavities were located. The lavage fluid was bloody (Fig. 2B), a bacterial culture was negative, and the results of an additional AFB smear and *M. tuberculosis* PCR were negative. The results of *Cytomegalovirus* (CMV) PCR, *Pneumocystis jirovecii* PCR, and *Aspergillus* antigen testing of the BAL fluid were positive.

On the fourth day of admission, the patient's blood pressure decreased with the onset of ventricular tachycardia on electrocardiography. His FiO_2 increased from 0.65 to 1.0 owing to increasing oxygen demand. On the eighth day of hospitalization, a cytologic examination of the BAL fluid confirmed the filariform larvae of *S. stercoralis* (Fig. 3), but the patient died on the day of the diagnosis, before

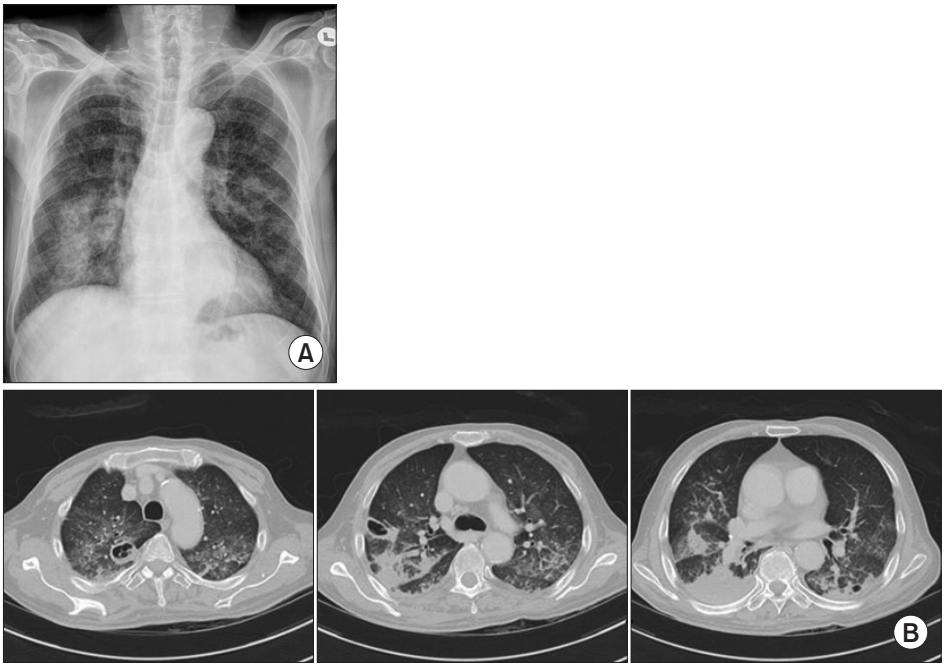

Fig. 1. (A) A chest radiograph shows bilateral and diffuse nodular infiltration and multiple patchy opacities in both whole lung fields, and a cavity in left hilar area. (B) Chest computed tomography scans reveal several thin-walled cavities in the right upper lobe and left lower lobe, and multiple consolidations, ground glass opacities and centrilobular nodules in both lungs.

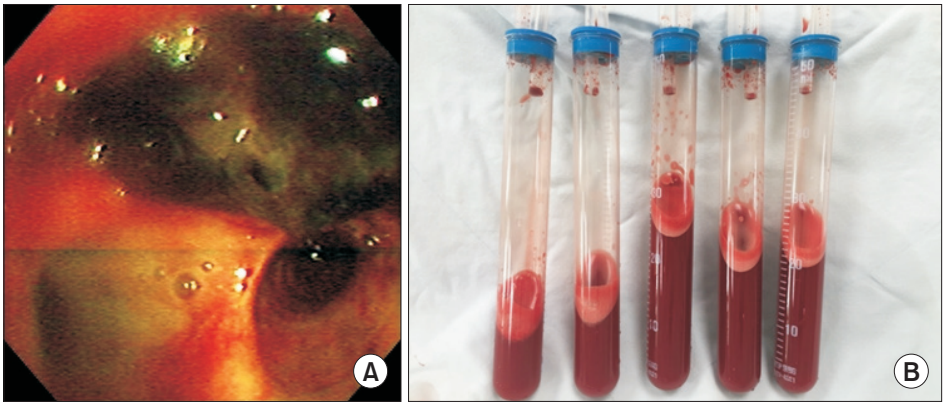

Fig. 2. (A) Bronchoscopy shows that the large blood clot plugged in the right upper lobe bronchus with hemorrhagic mucosa. (B) Bronchoalveolar lavage fluid shows bloody in appearance.

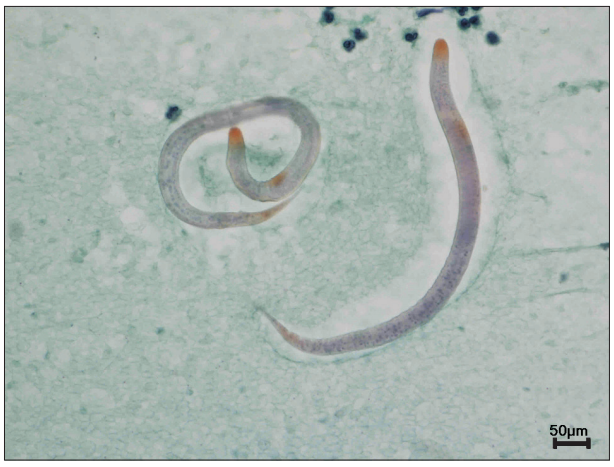

Fig. 3. Microscopic view of the bronchoalveolar lavage fluid shows filariform larvae of *Strongyloides* stercoralis (Gomori methenamine silver stain, ×400).

administration of the therapeutic agent.

DISCUSSION

Presented here is a case of *Strongyloides* hyperinfection presenting as ARDS and alveolar hemorrhage in an older patient in Korea with no immunosuppressive conditions.

Strongyloides hyperinfection is the term often used for the phenomenon in which the number of *S. stercoralis* organisms increases dramatically and worms are found especially in extraintestinal regions.[7]

S. stercoralis has the ability to reinfect within the human body, which is known as autoinfection, and it can live in the human body for extended periods of time, greatly increasing the parasite burden. Therefore, autoinfection can lead to hyperinfection, resulting in the development of complications involving the gastrointestinal tract, lungs, and peritoneum.[7,8]

Hyperinfection risk factors include decreased cellular immunity, age ≥65 years, chronic lung diseases, intestinal blind loops, achlorhydria, and the use of corticosteroids, antacids, or H2 blockers.[3,9] The use of corticosteroids was the main triggering factor that induced hyperinfection, but the cumulative dose or duration of the corticosteroid treatment causing *Strongyloides* hyperinfection was not known.[10] The patient in the present report had no history of systemic corticosteroid use, but he did undergo intra-articular corticosteroid injections. Intra-articular corticosteroids also have systemic side effects and may have been a risk factor for strongyloidiasis. In this case, I think that it was important to show that intra-articular corticosteroid injection could cause *Strongyloides* hyperinfection. Aging can be a risk factor as aging affects the function or number of T-cells.[9]

Peripheral eosinophilia may be absent in hyperinfection owing to the use of corticosteroids or the suppression of eosinophils due to bacterial infection.[9,11] Nabeya et al.[8] reported that there was no eosinophilic manifestation among 16 severe strongyloidiasis cases.

In hyperinfection, the larvae damage the intestinal mucosa and carry the enteric bacteria on their outer surfaces, resulting in bacterial and fungal infections often accompanied by leakage of gut flora.[7] Massive secondary infections are a major cause of death.[7] *Strongyloides* hyperinfection presented clinically with gram-negative sepsis in some reports.[12] CMV has been reported as a coinfection of *S. stercoralis* hyperinfection in cases of transplantation and HIV-1 infection.[13,14] In the present case, there was a coinfection with CMV, *P. jirovecii*, and *Aspergillus* spp., proven in the BAL fluid findings.

The patient presented here might have been exposed to *S. stercoralis* while walking barefoot in rice paddies, and the risk factors of older age and intermittent intra-articular administration of steroids may have led to *Strongyloides* hyperinfection. Given the presence of *P. jirovecii* and CMV as shown via PCR and *Aspergillus* antigen in BAL fluid, it is presumed that the vigorous movement of the larvae caused extensive bacterial, viral, and fungal infections, leading to the death of the patient.

In hyperinfection, respiratory specimens aid in the detection of parasites and larvae.[9] The prognosis varies depending on the severity of the strongyloidiasis, but the mortality rate exceeds 70%–80% despite appropriate treatment.[9]

Strongyloidiasis with acute respiratory failure and pulmonary hemorrhage is rare and almost is accompanied by an immunodeficiency disease.[5,6,8,12] In the present case, there was no definite underlying disease, but there was a history of intra-articular corticosteroid injection and old age. This is significant because *Strongyloides* hyperinfection may be associated with coinfection with CMV, *P. jirovecii*, and *Aspergillus* spp.

In Korea, the number of older patients with immunodeficiency due to the deterioration of biologic functions is increasing, and the chances of serious infections due to various parasites are increasing even in the absence of immunosuppressive disease.[15] Strongyloidiasis is difficult to diagnose because of the low parasite burden in Korea and the irregular larval output.[12] I report a case of *S. stercoralis* hyperinfection presenting as ARDS and alveolar hemorrhage in an older patient with no known immunosuppressive disease. In rare cases of ARDS accompanied by gastrointestinal symptoms in older patients, suspicion of parasitic infection and appropriate testing to confirm this suspicion may help to speed up the diagnosis and treatment of patients.

CONFLICTS OF INTEREST DISCLOSURES

The researcher claims no conflicts of interest.

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