Disseminated Strongyloidiasis in a patient with rheumatoid arthritis: A case report

Jin-Hao Zheng, Lu-Yu Xue

Abstract
BACKGROUND
Strongyloidiasis is usually a chronic infection, but it can develop into a fatal disease in immunosuppressed patients.

CASE SUMMARY
A 68-year-old male with rheumatoid arthritis was treated with a variety of immunosuppressants for the past 3 years. Recently, the patient appeared a partial small-bowel obstruction, petechia, coughing and peripheral neuropathy, diagnosis was difficult to clarify in other hospitals. Our hospital found S. stercoralis larvae with active movement in the stool routine and sputum smears. The diagnosis of disseminated strongyloidiasis was established. Ivermectin combined with albendazole was used for treatment. The patient was discharged in a healing condition.

CONCLUSION
This case underscores the importance of comprehensive differential diagnosis in immunocompromised patients.

Key Words: Strongyloidiasis; rheumatoid arthritis; immunosuppressants; small-bowel obstruction; ivermectin; albendazole

Core Tip: Strongyloidiasis is usually a chronic infection, but it can develop into a fatal disease in immunosuppressed patients. Here, we present a case of an immunocompromised patient with disseminated strongyloidiasis that was ignored by other hospitals. We discuss the challenges of diagnosis and treatment. As the disease was widespread, ivermectin combined with albendazole was used for treatment. This case underscores the importance of comprehensive differential diagnosis in immunocompromised patients.
INTRODUCTION

Strongyloidiasis is a disease caused by the human pathogenic parasitic roundworm *Strongyloides stercoralis*. Most larvae are excreted in the stool, but re-infection, or self-infection, can occur when the mature larvae burrow into the intestinal wall or the anal tissue. *S. stercoralis* infections can become chronic and even fatal in immunosuppressed patients. This report describes the clinical features of disseminated strongyloidiasis in an immunosuppressed patient, as well as the diagnosis and treatment.

CASE PRESENTATION

Chief complaints

A 68-year-old male repeated multi-joint pain for 3 years, abdominal pain and abdominal distension for 2 mo and progressive difficulty in swallowing, coughing, hoarseness, and dysphonia for 1 wk.

History of present illness

The patient with rheumatoid arthritis was treated successively using a variety of immunosuppressants (methylprednisolone, tocilizumab, adalimumab, rituximab) for the past 3 years. Recently, the patient received treatment in several hospitals for a partial small-bowel obstruction of unknown origin, which reoccurred repeatedly after treatment. As the patient’s condition worsened, new symptoms appeared, including petechia, progressive difficulty in swallowing, coughing, hoarseness, and dysphonia. A neurologist considered peripheral neuropathy because electromyography indicated peripheral nerve axonal damage. High-dose intravenous immunoglobulin therapy (2 g/kg over 5 days) was not effective, so plasmapheresis was recommended. At the same time, a parasite was detected in the stool, but the species was neither identified nor treated. Due to the progress of bulbar palsy, the patient was referred to our hospital.

History of past illness
Diabetic history: Diabetic history for several years, maximum 18mmol/L. Taking insulin medication, blood sugar unsatisfactory control.

**Personal and family history**
The patient had no specific personal and family history.

**Physical examination**
Upon admission, he displayed weight loss, stable vital signs, hoarseness, dysarthria, wet rales audible in both lungs, weak bowel sounds, muscle strength grade 3 in all limbs, and diminished tendon reflexes.

**Laboratory examinations**
His biochemistry panel was as follows: K 3.4 mmol/L, Na 129 mmol/L, Ca 1.88 mmol/L, and albumin 25 g/L. Stool-Rt and sputum smears tested positive for *S. stercoralis* larvae with active movement.

**Imaging examinations**
A chest CT showed bilateral infiltrates indicating pneumonia. Echocardiography showed impaired movement of the left ventricular myocardium (EF 42%).

**FINAL DIAGNOSIS**
The diagnosis of disseminated strongyloidiasis was established.

**TREATMENT**
After 1 wk of treatment with albendazole 400 mg tid and other supportive treatments, the sputum smear was still positive, then ivermectin 0.2 mg/kg/d +2 days every 2 wk was given. On day 4 of ivermectin treatment, the sputum smear and stool tested negative for intestinal parasites. After 2 wk of comprehensive treatment, the patient's mental state gradually improved and muscle strength of the limbs recovered. After 6
wk of hospitalization, his abdominal pain and all previously mentioned symptoms except for the joint pain healed. The patient was discharged and given a small dose of methylprednisolone + methotrexate + celecoxib to control the rheumatoid arthritis and relieve the joint pain. We used albendazole for 4 wk totally and ivermectin for 6 wk totally\(^2\).

**OUTCOME AND FOLLOW-UP**
At 3 mo after discharge, a follow-up chest CT and electromyography showed lung and cardiac function had recovered.

**DISCUSSION**
Strongyloidiasis is a zoonotic intestinal parasitosis caused by *S. stercoralis*. It is estimated that 30–100 million people are infected worldwide with this parasite\(^3\). Most infected individuals are asymptomatic or present with intermittent symptoms\(^4\). Immunosuppressed patients can develop hyperinfection syndrome and disseminated nematode disease, which have high mortality rates\(^5\)\(^,\)\(^7\). Strongyloidiasis has been reported following concomitant tocilizumab and methylprednisolone treatment\(^8\). Some case reports suggest that paralytic ileus may be caused by massive intestinal infestation with *S. stercoralis*\(^9\).

Our case has two important clinical features. First, the patient had a history of immunosuppression and subsequently developed clinical symptoms (e.g., intestinal obstruction, pneumonia, and petechia). The patient’s heart also was affected. Previous hospitals detected the presence of parasites but focused instead on the neurological manifestations. We confirmed the presence of *S. stercoralis* larvae in the patient’s stool and sputum\(^10\). Second, the patient presented with choking and hoarseness at the time of diagnosis. Head, neck, mediastinal MRI, cerebrospinal fluid, and other examinations found no evidence of neurological invasion. Therefore, we considered two possibilities: 1) nutritional deficiencies in vitamin B1, vitamin B12, and folic acid due to long periods of fasting, causing malabsorption and intestinal obstruction, which can lead to
peripheral neuropathy[11]; 2) neurotoxic biological agents (e.g., TNF inhibitors, anti-IL-6 receptor antibody), which can cause peripheral neuropathy in approximately 42% of cases[12]. The Centers for Disease Control and Prevention and World Health Organization recommend ivermectin as the first choice for strongyloidiasis. In endemic areas, a combination of albendazole and ivermectin is recommended[13], and Moxidectin has also been tried as a treatment[14]. Repeated or extended dosing is preferred until worms are no longer detected[15]. Considering that the patient was still taking low-dose methylprednisolone and methotrexate tablets for rheumatoid arthritis, we adopted a multi-dose and long course of treatment. At the 3-month follow-up, no recurrence of the disease was detected, so the treatment was effective.

CONCLUSION

This case highlights important considerations for patients receiving immunosuppressive therapy. It is necessary to improve medical workers’ awareness of strongyloidiasis to avoid delays in diagnosis and ensure adequate management of infected patients.
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