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ABOUT COVER

Editorial Board Member of *World Journal of Clinical Cases*, Cristina Tudoran, PhD, Assistant Professor, Department VII, Internal Medicine II, Discipline of Cardiology, "Victor Babes" University of Medicine and Pharmacy Timisoara, Timisoara 300041, Timis, Romania. cristina13.tudoran@gmail.com

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

Jin-Hao Zheng, Lu-Yu Xue

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Jin-Hao Zheng, Lu-Yu Xue, Department of Critical Care Medicine, Huashan Hospital Affiliated to Fudan University, Shanghai 200040, China

Corresponding author: Lu-Yu Xue, Doctor, Occupational Physician, Department of Critical Care Medicine, Huashan Hospital Affiliated to Fudan University, No. 12 Middle Urumqi Road, Shanghai 200040, China. icuxueluyu@163.com

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 presented with a partial small-bowel obstruction, petechia, coughing and peripheral neuropathy. The diagnosis was difficult to clarify in other hospitals. Our hospital found *Strongyloides stercoralis* larvae with active movement in the routine stool and sputum smears. The diagnosis of disseminated strongyloidiasis was established. Ivermectin combined with albendazole was used for treatment. The patient responded to therapy and was discharged.

CONCLUSION

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

Key Words: Strongyloidiasis; Rheumatoid arthritis; Immunosuppressants; Small-bowel obstruction; Ivermectin; Albendazole; Case report

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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 the treatment. Since the disease was widespread, ivermectin combined with albendazole was used for treatment. This case underscores the importance of comprehensive differential diagnosis in immunocompromised patients.

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INTRODUCTION

Strongyloidiasis is a disease caused by the human pathogenic parasitic roundworm *Strongyloides stercoralis* (*S. 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[1]. 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 with 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 d) was not effective, so plasmapheresis was recommended. At the same time, a parasite was detected in the stool, however, 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 18 mmol/L. Taking insulin medication, blood sugar is unsatisfactory for control.

Personal and family history

The patient had no specific personal and family history.

Physical examination

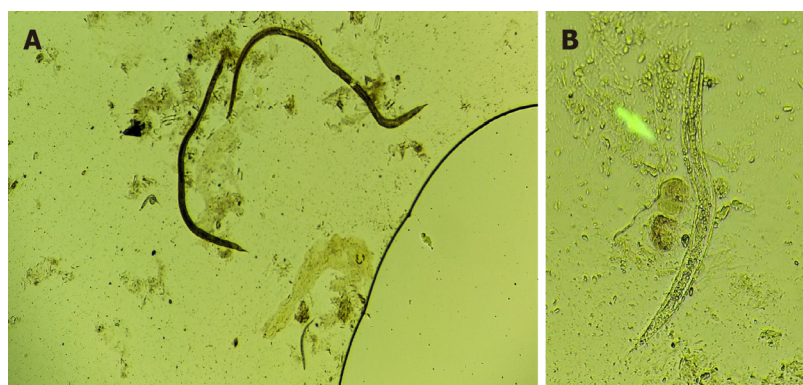
Upon admission, he displayed weight loss, stable vital signs, hoarseness, dysarthria, wet rales audible in both lungs, weak bowel sounds, muscle strength grade 3 in all limbs and diminished tendon reflexes.

Laboratory examinations

His biochemistry panel was as follows: K of 3.4 mmol/L, Na of 129 mmol/L, Ca of 1.88 mmol/L, and albumin of 25 g/L. Stool-Rt and sputum smears tested positive for *S. stercoralis* larvae with active movement (Figure 1).

Imaging examinations

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



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Figure 1 Larva of *Strongyloides stercoralis* separated from the stool of the patient. A: 10 × 10; B: 10 × 40.

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. The addition of ivermectin 0.2 mg/kg/d × 2 d every 2 wk was then 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 had dissipated. 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 total and ivermectin for a total of 6 wk[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 was also 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]; and (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 mo 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.

FOOTNOTES

Author contributions: Zheng JH designed the study, analyzed the data and wrote the manuscript; Xue LY contributed to study conception and design and revision of the manuscript.

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Country/Territory of origin: China

ORCID number: Jin-Hao Zheng 0000-0003-0864-5156; Lu-Yu Xue 0000-0001-8773-7088.

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