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A case of triple infection including strongyloides stercoralis in a microscopic polyangiitis patient

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ABSTRACT

We present the case of a microscopic polyangiitis (MPA) patient who developed strongyloidiasis, *nocardia and citrobacter freundii* (CF) infection after corticosteroides and immunosuppressant therapy. When digestive, respiratory or other system symptoms consecutively occur in a immunocompromised host who lives in tropical or temperate zone and have close contact with soil, we should take strongyloidiasis into consideration despite absence of eosinophilia. Mixed infection with *nocardia* cannot be easily excluded. It is essential to search for the etiology proof with multiple approaches positively and repeatedly.

1. Introduction

Strongyloidiasis is a soil-transmitted helminthosis caused by *strongyloides stercoralis,* which is easily to be overlooked among tropical diseases resulting from the lack of specific clinical manifestations. Many people infected with *S.stercoralis* are prone to developing chronic infection but unware of it due to its special auto-infection cycle and indefinite life span in the human host [1]. As the immunosuppressed population continues to increase in recent years, Strongyloidiasis has gradually evolved as an emerging disease, which could turn into extreme dissemination with fatal outcome.

2. Case presentation

A 58-year-old male farmer, local resident, who had been diagnosed as MPA, received intravenous cyclophosphamide (500mg) semimonthly and oral prednisolone (40mg/d) for two months and discharged after improvement, was readmitted due to hyperpyrexia, ventosity, hypodynamia and inappetence for a week. Initial vital signs at his second hospitalization were temperature 38.0 °C, blood pressure 104/66 mmHg, heart rate 102 beats/min, respiration rate 20/min, oxygen saturation 95 %.There was no conspicuous abnormality in general physical examination. Blood routine results were as follows: white blood cells 6.7*10^9/L, Neutrophils 79 %, eosinophils 3%, hemoglobin 70g/l and normal platelets. Hypoproteinemia (22.4g/l), hyponatremia (129mmol/l) and elevated C -reactive protein (43.2mg/l) were observed. Liver function was almost normal except for transaminase elevated slightly. Kidney function, HIV antibody, arterial blood gas analysis, rheumatism, tuberculosis and virus related indicators as well as cranial magnetic resonance imaging were all basically normal. Computed tomography scan revealed high density patchy shadow in right superior lung lobe, a nodular cavity in left lower lung lobe and focal small intestine wall thickening. (Figs. 1 and 2).

After intravenous of Sulperazone for three days, the farmer's body temperature dropped to normal. However, his condition didn't improved. Instead, he appeared abdominal pain, anus without defecating, dyspnea, cough and sputum. Expectant treatment resulted in defecation and pain relief. Meanwhile, bedside bronchoscope was performed. A mass of *S. stercoralis, nocardia and citrobacter freundii* (CF) were found both in sputum and bronchoalveolar lavage fluid (BALF) on day 5 of hospitalization. (Figs. 3 and 4). Hence, we adjusted the therapeutic regimen to Albendazole (400mg/d), Tienam (0.5g, ivgtt, q8h), and Cotrimoxazole (0.48,po.tid). Unfortunately, two days later, the patient quickly had a fever again, developed severe septic shock, obstinate respiratory failure, and finally died.

3. Discussion

Approximately 370 million people have infected S.stercoralis worldwide, mostly in tropical and subtropical zones [2]. Patients are the prime source of infection, who could continuously have been infected for many years without overt symptoms even after leaving endemic

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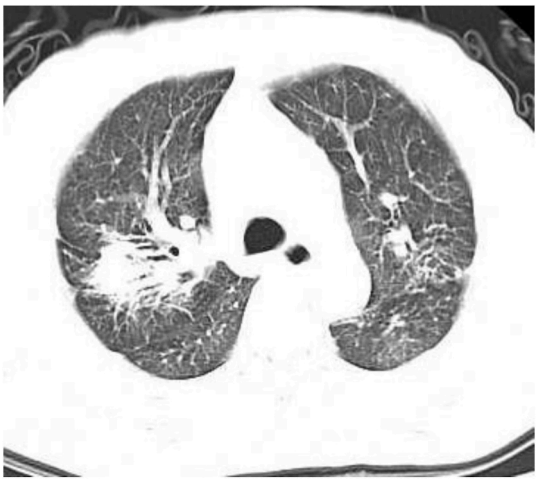

Fig. 1. High density patchy shadow in right superior lung lobe.

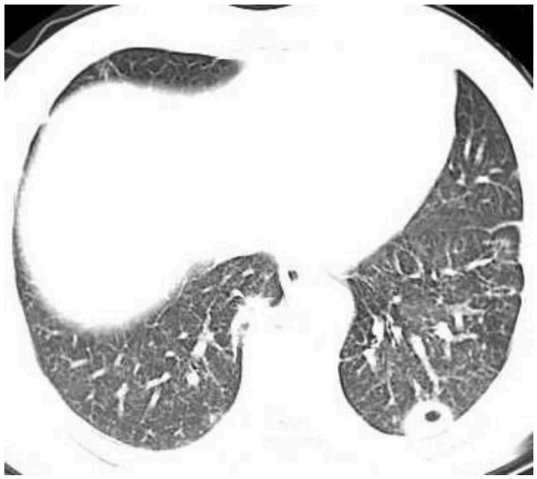

Fig. 2. A nodular cavity in left lower lung lobe.

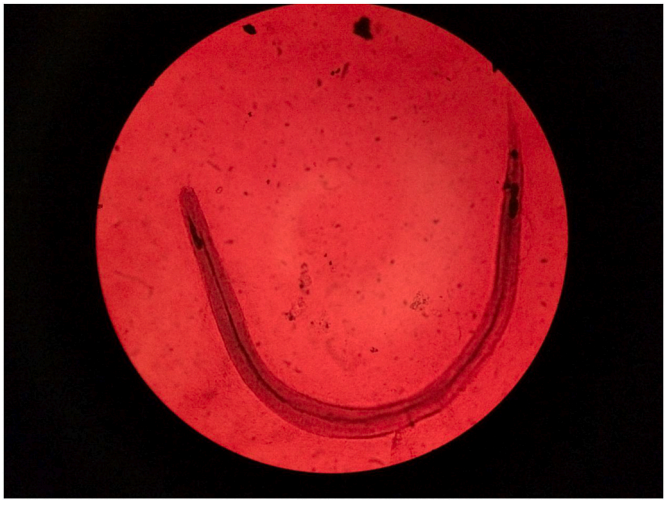

Fig. 3. Iodine liquid dye: filariform larvae of *S. stercoralis* × 1000.

areas. In China, most cases were reported in southern provinces, such as Guangdong, Guangxi, Taiwan and so on. People living in rural or suburban areas with poor sanitation, usually acquired the disease through

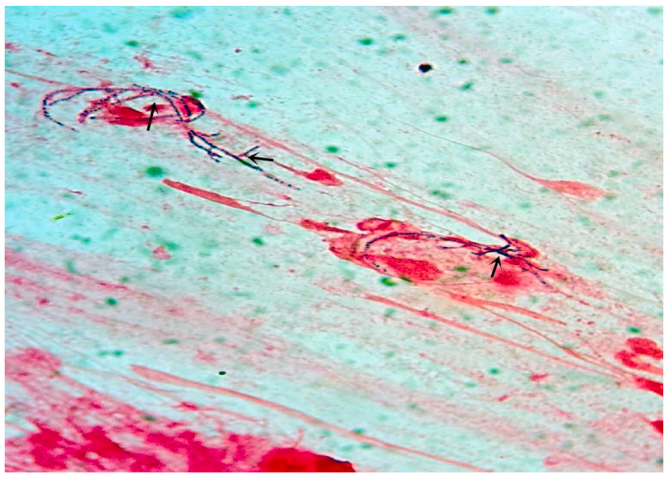

Fig. 4. Gram staining: mottled with 90° branching hyphae.(*Nocardia*) × 1000.

eating contaminated food or close contact with soil in work and life [3]. In our case, the patient was a native farmer who had never been to other places beyond Jiangxi province, located in the southeastern China, where the climate is relatively warm.

Human immunodeficiency virus (HIV) infection, human T-lymphotropic virus type 1(HTLV-1) infection, alcoholism, malignancy and immunocompromised state are main high risk factors [4]. Prednisone treatment at 20mg/d for two weeks is considered as a high risk factor for the development of strongyloides stercoralis hyper-infection syndrome [1]. The pathogenicity degree of strongyloidiasis varies depending on the body's immune state. Once the soil-transmitted helminth infection occurs in a immunodeficient host, it is often misdiagnosed owing to various systems involved, and prone to fatal outcome, with up to 90 % mortality [2].

Peripheral blood eosinophil elevation is common in parasitic infection. As part of the innate immunity, eosinophils play a prominent role in killing worms and triggering Th2 immune responses as antigen-presenting cells in strongyloidiasis [5]. But in this case, no eosinophil increase was observed throughout the whole course, which led us to fail to take parasitic infection into account at the initial stage. Hays concluded that eosinophils had only sensitivity 60.9 % and specificity of 71.1 % [6]. Wurtz R found that peripheral blood eosinophils were lower in the death group than in the non–death goup, which suggested that normal or hypoeosinphils in strongyloidiasis patients may mean a poor prognosis. This may be related to the increase of cell apoptosis of Th2 caused by eosinophils decrease [7]. Pang reported a case that eosinophil elevated from 3.9 % to 10 % after discontinuation of steroids [8]. It maybe because corticosteroids or immunosuppressants could impair even inhibit the eosinophilic immune responses.

The patient appeared digestive symptoms; abdominal CT displayed regional small intestinal wall thickening, but we failed to identify whether the S.stercoralis caused digestive tract infection since the patient's conditon progressed too fast to perform gastrointestinal endoscopy. Our case showed negative results of stool tests on account of intermittent excretion of lavae, and lower detection rate of direct smear method [9].

As a clinical common enterobacteria, CF, which is an opportunistic pathogen, could cause septicemia, diarrhea, meningitis, urinary tract and respiratory infection when individuals immunity decline [10]. Mechanical irritation and toxic effect from S.stercoralis can induce gastrointestinal lesions and translocation of intestinal flora, which facilitate enterobacteriaceae infection [11].

Nocardia exists widely in nature. People could acquire nocardiosis when respiratory tract, broken skin or alimentary canal comes into contact with the contaminated soil, dust or food. Because of indistinguishable clinical and imaging manifestations as well as lower positive

rate of sputum culture in nocardiosis, many cases remain underdiagnosed. The prognosis of pulmonary nocardiosis was particularly poor in patients treated with immunosuppressants. Furthermore, pulmonary nocardiosis induces mixed or disseminated infections at a high frequency. The key diagnosis requires positive specimen culture or PCR [12].

This case is a complicated triple infection including S.stercoralis,C. freundii and nocardia.Similar cases have rarely been reported. The patient was a farmer who was using hormone and immunosuppressants and was at risk of exposure to contaminated soil. Both S.stercoralis and nocardia infection can be acquired by broken skin contacting with contaminated soil. And strongyloidiasis could trigger enterobacterial infection. Although we adjusted antibiotic regimen as soon as the pathogen was established, the patient died of a critical illness.

4. Conclusion

To avoid S.stercoralis infection, people should develop good personal hygiene habits. Prior to the use of glucocorticoid or other immunosuppressants, screening for serological tests of S.stercoralis ought to be performed in high risk group. Farmers who take hormones or immunosuppressants and live in tropical or temperate country or region, should take personal precautions, such as wearing rain boots and gloves when in contact with soil. Once immunodeficient patients are clinically suspected of strongyloidiasis, although peripheral blood eosinophils count is not high or even low, stool and serological test, relevant endoscopy and PCR examinations should be actively conducted to obtain pathogenic diagnosis as soon as possible [13].

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Declaration of competing interest

All the authors have seen and approved the manuscript. The authors report no conflicts of interest.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.rmcr.2021.101479.

Patient consent

Written informed consent was obtained from the patient's family member for the publication of this manuscript in *Respiratory Medicine Case Reports*.

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