Disseminated *Strongyloides stercoralis*: Hyperinfection during medical immunosuppression

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Hyperinfection caused by *Strongyloides stercoralis* in iatrogenically immunosuppressed patients is becoming more frequently observed. Here, we review the relevant literature and present a recent case of hyperinfection syndrome of *S stercoralis* in a patient chronically treated with systemic corticosteroids and methotrexate for dermatomyositis. The patient was born in Guatemala but no history of *Strongyloides* infection was documented. Disseminated *Strongyloides* is often associated with the immunocompromised state and is commonly seen with cutaneous lesions, respiratory failure, and sepsis. In this patient, a protracted course of progressive muscle weakness and multiple hospital stays for respiratory distress led to acute respiratory failure, septic shock, and rapid physical decline. A few days preceding his death, the patient developed petechiae and multiple purpuric macules and patches over the abdomen and thighs. Histologic review of skin biopsy specimens demonstrated multiple intravascular and interstitial filariform larvae. Dermatologists should be aware of patient populations at risk for infection with *S stercoralis* and be able to make this diagnosis to initiate earlier treatment of hyperinfection and dissemination. (J Am Acad Dermatol 2010;63:896-902.)

Key words: dermatomyositis; dissemination; hyperinfection; immunosuppression; parasites; *Strongyloides stercoralis*.

Reports highlighting the diagnostic challenges of *Strongyloides stercoralis* hyperinfection[1] underscore the need for improved recognition of the multiple clinical presentations of parasite infections. This is particularly troublesome in nonendemic areas[2,3] and among US physicians in general.[4] Many cases of strongyloidiasis in the United States are misdiagnosed[4] as pneumonia,[5,6] inflammatory bowel disease,[7-9] and systemic lupus erythematosus.[10] Cutaneous presentations have been misdiagnosed as lichen simplex chronicus[11] and a drug eruption.[12] Many deaths associated with disseminated *Strongyloides* are considered to be secondary to poor screening among patients at risk for disease.[4]

S stercoralis is an intestinal nematode endemic to tropical and subtropical areas of the world and temperate areas such as the southeastern United States.[13-15] Immunocompetent hosts infected by the parasite can be asymptomatic for decades. However, common presenting symptoms of acute disease and hyperinfection include abdominal pain and respiratory difficulty.[16] Dissemination leading to sepsis as a result of enteric pathogens,[17,18] cutaneous lesions,[19-21] neurologic involvement, and respiratory failure is often observed in carriers who become immunocompromised.[16] Although there are prior reports of *Strongyloides* hyperinfection in patients with a variety of autoimmune diseases, an association with dermatomyositis has not been previously been reported.

CASE REPORT

A 39-year-old man who immigrated to the United States from Guatemala with a history of dermatomyositis on chronic immunosuppressive therapy presented with shortness of breath, abdominal pain, nausea, and vomiting for 1 day. In the emergency department the patient was febrile, hypotensive, and tachycardic. Empiric treatment of sepsis was initiated

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Funding sources: None.
Conflicts of interest: None declared.

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Published online February 22, 2010.
0190-9622/$36.00

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doi:10.1016/j.jaad.2009.09.037

in addition to intravenous hydrocortisone every 8 hours. The patient was moved to the intensive care department where he was intubated for respiratory distress. Notably, the patient had been previously placed on nightly ventilator support after an extended hospitalization for *Acinetobacter* pneumonia.

The patient's medical history was significant for approximately 1 year of progressively worsening proximal muscle weakness. A diagnosis of dermatomyositis was made based on the patient's proximal muscle weakness, elevated creatine kinase level, muscle biopsy specimen confirming myositis, electromyogram results, and a heliotropic rash. Over the course of months, the patient was treated on several occasions for severe respiratory distress that was thought to be secondary to dermatomyositis associated diaphragmatic weakening. Leading up to the current hospitalization, the patient underwent many weeks of physical therapy and multiple medical treatments including: methylpredisolone, a trial of rituximab, intravenous immunoglobulin, and weekly methotrexate injections. His daily medical regimen on admission included prednisone (45 mg daily), methotrexate (2.5 mg/wk), leucovorin (5 mg once/wk), and dapsone (100 mg twice daily).

Imaging and laboratory results revealed acute pathology of the small bowel and lungs. Computed tomography of the chest and abdomen revealed bilateral lung consolidations, marked small bowel dilatation with air fluid levels, and free fluid in the pelvis. The source of pneumonia was thought to be secondary to aspiration in the context of an ileus. Blood cultures grew *Escherichia coli* and *Klebsiella*; sputum samples grew *E coli* and *Klebsiella* in addition to *Acinetobacter* and *Proteus*. The patient's antibiotic regimen was adjusted appropriately. Over the course of the next week the patient's clinical state continued to decline.

On day 9, the patient developed multiple, purpuric macules and patches on his abdomen and proximal thighs (Fig 1). Bronchoscopy samples demonstrated parasites consistent with *S stercoralis* infection (Fig 2). Simultaneously, histologic review of a biopsy specimen from the patient's abdomen revealed leukocytoclastic vasculitis and multiple filariform larvae found intravascularly and interstitially throughout the dermis (Fig 3). A serum IgG enzyme-linked immunosorbent assay (ELISA) for *S stercoralis* revealed undetectable antibody levels and the patient's blood eosinophil levels were within normal limits throughout his hospital stay. However, because of the patient's deteriorating clinical symptoms and his laboratory and histologic findings, albendazole (400 mg twice daily), ivermectin (12 mg daily), and fluconazole (400 mg daily) were started.

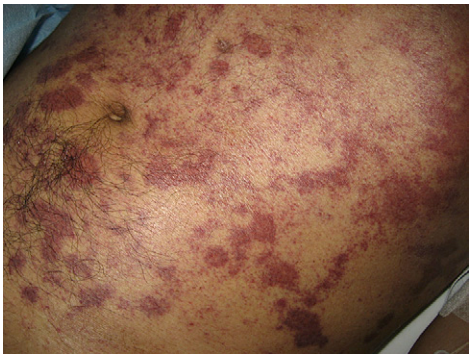

Fig 1. Petechiae and purpuric macules and patches developed on patient's abdomen and thighs.

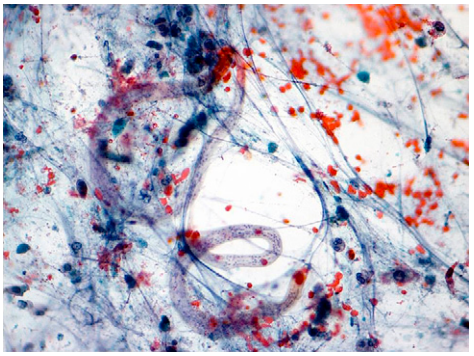

Fig 2. Bronchial lavage samples revealed larvae consistent with *Strongyloides stercoralis*.

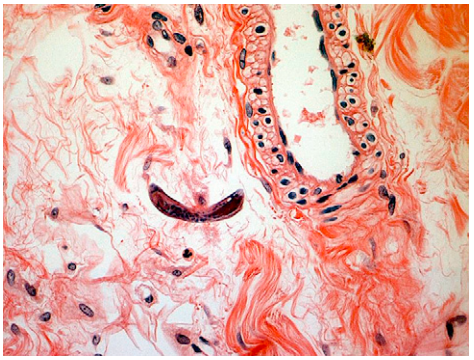

Fig 3. Skin biopsy specimen with mild superficial and deep perivascular infiltrate composed of neutrophils, leukocytoclasis, and presence of parasite larvae. Morphology of skin larva is consistent with *Strongyloides*.

Over hospital days 10 to 12 the patient became hemodynamically unstable and was unresponsive to pressor support. On hospital day 12, the patient died.

At autopsy, the patient was found to have multiple purpuric macules of the trunk and lower extremities and generalized muscle atrophy. Gross examination of the organs demonstrated moderate cerebral edema with congested vessels, bilateral pulmonary hemorrhage, fatty liver changes, and edematous

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