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ATYPICAL CLINICAL PRESENTATION OF STRONGYLOIDIASIS IN A PATIENT CO-INFECTED WITH HUMAN T CELL LYMPHOTROPHIC VIRUS TYPE I

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Abstract. Alterations in the immunologic response induced by human T cell lymphotrophic virus type I (HTLV-I) predispose the development of disseminated strongyloidiasis. We report a case of an atypical clinical presentation of strongyloidiasis in a patient co-infected with HTLV-I causing scrotal and perineal pain and infertility. *Strongyloides stercoralis* was found in the analysis of the sperm and specific therapy for strongyloidiasis was associated with disappearance of the symptoms.

INTRODUCTION

Strongyloidiasis has a worldwide distribution and is one of the most important enteric helminthic infections. Diarrhea, abdominal pain, and less frequently vomiting are the main clinical features of the disease. More recently, an association between human T cell lymphotrophic virus type I (HTLV-I) and disseminated strongyloidiasis has been reported.[1–4] This association is due to decreased immunity against *Strongyloides stercoralis* in patients co-infected with HTLV-I. The high production of interferon-γ (IFN-γ) resulting from HTLV-I infection leads to a decreased synthesis of interleukin-4 (IL-4), IL-5, parasite-specific IgE, and eosinophils, cytokines and cells that are involved in the expulsion and killing of *S. stercoralis*, respectively.[5–7] Herein, we report a patient co-infected with HTLV-I and *S. stercoralis* who sought medical evaluation for infertility. Upon evaluation, *S. stercoralis* was detected in the ejaculate mixed with spermatozoa and in the urine.

CASE HISTORY

A 27-year-old man born in Salvador, Bahia, Brazil had sperm and urine analyses performed to evaluate infertility. On clinical evaluation, the patient complained of mild pain in the scrotum and perineal region. He denied having diarrhea, abdominal pain, or vomiting. There was mild edema in the scrotum and a urologic examination found small varicoceles. An ultrasound examination showed no abnormalities in the abdomen, but a scrotal examination confirmed the clinical finding of varicoceles. A spermatogram showed a normal number of spermatozoae with normal motility and a large number of rhabditiform and filariform larvae and young adult female *S. stercoralis*. Contamination of the sperm by urine was ruled out by the aspect of the material and by the forms of parasite found. While rhabditiform larvae were detected only in the urine, all parasite stages were observed in the sperm. The total peripheral white blood cell count and number of eosinophils were normal. Rhabditiform larvae of *S. stercoralis* were also observed by urine analysis and in the stool. Because of the atypical presentation of *S. stercoralis* infection, HTLV-I serology was performed. A diagnosis of HTLV-I infection was made and confirmed by Western blot.[7] Cytokines were measured in supernatants of mononuclear cells stimulated and not stimulated with *S. stercoralis* antigen. The levels of IFN-γ, tumor necrosis factor-α, IL-5, and IL-13 (Table 1) were measured by an enzyme-linked immunosorbent assay sandwich technique (Genzyme Corp., Cambridge, MA) and the results were expressed in picograms per milliliter based on a standard curve generated using recombinant cytokines.

The patient was treated with a single dose of cambendazole (5 mg/kg of body weight). The pain in the scrotum and perineal region and edema resolved and the varicoceles were reduced in size. *Strongyloides stercoralis* was no longer seen on a spermatogram, but rhabditiform larvae were present in the stool examined two months after therapy. The patient was re-treated with a single dose of ivermectin (200 μg/kg of body weight) and the results of a repeated spermatogram and stool examinations every 2–3 months remained negative for *S. stercoralis* at one year of follow-up. After therapy, his wife became pregnant, indicating that the infertility had improved.

DISCUSSION

Disseminated *S. stercoralis* infection is a recognized consequence of immune suppression caused by corticosteroids or cytotoxic drugs.[8,9] More recently, severe and recurrent strongyloidiasis has been reported in association with HTLV-I co-infection.[1–4,10–12] In disseminated strongyloidiasis, increases in parasite load from autoinfection leads to systemic migration of *S. stercoralis* to other organs. The lungs, liver, and brain are the most common organs invaded by *S. stercoralis*, leading to severe clinical disease. Herein, we described a patient who complained of scrotal and perineal pain in whom *S. stercoralis* was found in a spermogram. The documentation of *S. stercoralis* larvae and adult worm in the sperm and the improvement of perineal symptoms and signs after specific anti-strongyloides treatment indicated not only the ability of this parasite to infect the genitourinary tract, but

TABLE 1

Levels of IFN-γ, TNF-α, IL-5, and IL-13 in supernatants of lymphocyte cultures stimulated with *Strongyloides stercoralis* antigen*

	Levels (pg/ml)	
Cytokines	Medium	*S. stercoralis* antigen
IFN-γ	70	246
TNF-α	0	0
IL-5	185	184
IL-13	207	191

* Peripheral blood mononuclear cells were stimulated with 5 μg/mL of *S. stercoralis* antigen as previously described.[5] IFN-γ = interferon-γ; TNF-α = tumor necrosis factor-α; IL-5 = interleukin-5.

also to cause symptoms. Patients co-infected with HTLV-I and *S. stercoralis*, as observed in this case, have higher levels of IFN-γ and lower levels of IL-5 and IL-13 than patients who are infected with *S. stercoralis* alone.[6] The higher IFN-γ production by lymphocytes in patients infected with HTVL-I and *S. stercoralis* leads to a decreased *S. stercoralis*-specific type 2 immune response characterized by low levels of IL-4, IL-5, and IL-13,[5,6] cytokines that are critical for the control of *S. stercoralis* infection. Clinically, patients co-infected with HTLV-I and *S. stercoralis* may develop severe strongyloidiasis, as well as a low cure rate in response to anti-*S. stercoralis* drugs.[13] This report calls attention to an atypical clinical presentation of *S. stercoralis* in a patient co-infected with HTLV-I in whom the clinical manifestation was related to the dissemination of *S. stercoralis* into the genitourinary tract, resulting in infertility and scrotal and perineal pain.

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