Case Report: Myocarditis during Acute Schistosomiasis in Two Travelers

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Abstract. We report two cases of myocarditis complicating acute schistosomiasis in returning travelers. Treatment with corticosteroids led to full recovery in both cases. Although the pathophysiology of this complication remains unclear, we recommend treating such patients with corticosteroids rather than praziquantel, which can be associated with clinical deterioration.

INTRODUCTION

Schistosomiasis is a helminthic disease of major importance on a worldwide scale. Acute (or invasive) schistosomiasis (AS) is especially observed in nonimmune travelers returning from endemic areas.1 It is usually characterized by a benign clinical course but life-threatening events such as neurological complications can occur.2,3 Cardiac events have been rarely reported during AS but may also be potentially lethal. We report two cases of myocarditis complicating AS.

CASE REPORTS

The first patient was a 16-year-old French tourist who had been traveling for 1 month in West Africa, Burkina Faso, and Mali. During his journey, he bathed once in a waterfall in the Dogon area of Mali. Four weeks later, he was hospitalized in Paris for fever (39–39.2°C) and arthromyalgias lasting for 10 days. He presented with urticarial rash, cough, dyspnea, face edema, and oppressive retrosternal chest pain for 2 days. These symptoms appeared 26 days after the exposure. His eosinophil count was 2,000/mm3 (22%), and an acute nonspecific repolarization disorders: inverted ST-waves in lead V1 and depression of the S-T segment in lead V4, V5, V6 and DII (Figure 1). The troponin level was 13.9 µg/L (normal < 0.2 µg/L) and the creatine kinase-MB form (CK-MB) level was 6.6 µg/L (normal < 5 µg/L). Results of echocardiography were normal. Aspartate and alanine aminotransferase levels were 115 IU/L and 225 IU/L, respectively. Multiple stool and urine samples were negative for parasite eggs, larvae, and adult forms of schistosomes. Serodiagnosis of schistosomiasis was positive (1/800) by indirect immunofluorescence (S. mansoni in-house prepared antigen; Laboratoire de Parasitologie-Mycologie, Groupe Hospitalier Pitié-Salpêtrière, Paris, France) and by hemagglutination (Cellognost Schistosomiasis; Chiron-Behring, Marburg, Germany). AS was suspected. Electrocardiogram showed nonspecific repolarization disorders in apico-lateral territory: negative T-wave in lead V1 and depression of the S-T segment in lead V4, V5, V6 and DII (Figure 1). He was thus admitted in the Cardiological Intensive Care Unit. Results of echocardiography showed segmentary hypokinesia (apical and anterior medium third). Troponin levels rose to 4.5 µg/L. The patient received intravenous corticosteroids (prednisone 1 mg/kg/d) and acebutolol associated with ramipril for acute myocarditis. The cardiac magnetic resonance imaging (MRI) was normal. Corticosteroid therapy was gradually reduced. Three months later, he received praziquantel (Biltricide®, Bayer Santé, Loos, France) (40 mg/kg). In the following months, clinical examinations, eosinophil count, and troponin remained normal. One year later, the patient was clinically well.

The second patient was a 21-year-old French traveler who was hospitalized for persistent fever (38–39°C), myalgias, headache, dry cough, and loss of appetite. One month earlier, he had developed a maculopapular rash just after bathing once, with other members of a tourist group, in a lake in the Dogon area of Mali where he had been traveling for one month. The first clinical sign was a self-limited right palpebral angioedema that appeared 27 days after the exposure, as well as swimmer’s itch. His eosinophil count was 2,000/mm3 (22%), and an acute schistosomiasis was suspected. Electrocardiogram showed nonspecific repolarisation disorders: inverted ST-waves in lead V4 (Figure 1). The troponin level was 13.9 µg/L (normal < 0.2 µg/L) and the creatine kinase-MB form (CK-MB) level was 6.6 µg/L (normal < 5 µg/L). Results of echocardiography were normal. Aspartate and alanine aminotransferase levels were 115 IU/L and 225 IU/L, respectively. Multiple stool and urine samples were negative for parasite eggs, larvae, and adult forms of schistosomiasis. Serodiagnosis of schistosomiasis, initially negative, became positive by indirect immunofluorescence (1/800) (S. mansoni in-house prepared antigen; Laboratoire de Parasitologie-Mycologie, Groupe Hospitalier Pitié-Salpêtrière, Paris, France) and by hemagglutination (1/512) (Cellognost Schistosomiasis; Chiron-Behring, Marburg, Germany). The patient received praziquantel (40 mg/kg). Two days later, he abruptly developed mental confusion, anosognosia, and splinter hemorrhages under his nails. His temperature was normal. Cerebral MRI showed multiple bilateral brain infarcts suggestive of cerebral vasculitis. Electrocardiography showed persistent nonspecific repolarisation disorders. Heart computed tomography was normal, with slight myocardium and dry pericardium. Corticosteroid therapy (prednisone 1 mg/kg/d) was started, and the symptoms disappeared within 48 hours. Eosinophil count fell down to 300/µm3 and troponin level became negative 2 weeks later. Cardiac MRI was performed 2 month later and revealed a discrete delayed myocardial perfusion of the septum with subendocardial enhancement compatible with endocardial fibrosis. Four months later, S. haematobium eggs were found by direct microscopic examination of centrifugated urine samples from the patient and five of his friends who had bathed in the same lake. In the next 2 years, clinical examinations, eosinophil count, and troponin levels were still normal.

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**DISCUSSION**

Acute schistosomiasis typically occurs in nonimmune persons 3 to 6 weeks after exposure during a bath in an endemic area. It is considered as a systemic hypersensitivity reaction against the migrating schistosomulae in tissue. It’s usually self-limited and subsides over a few weeks, although life-threatening complications have been described that were mainly neurological. In our two cases, the first signs of AS appeared 26 and 27 days after the exposure, respectively. Our two cases show that cardiac complications can be added to the clinical spectrum of AS. These cardiac signs differ from those observed during the chronic phase of schistosomiasis.

Our two patients presented with acute myocarditis during AS. In the first case, chest pain occurred at the same time of troponin level elevation and electrocardiogram abnormalities despite negative MRI. In the second case, the patient did not present any cardiac signs but the diagnosis of myocarditis relied on high troponin level and electrocardiogram repolarisation abnormalities. In both cases the patients were nonimmune travelers presenting with hypersensitivity symptoms and eosinophilia characteristics of AS. The involved species is *S. haematobium* according to the results of urinalysis in the second patient; the first patient bathed in the same area of Mali.

The involvement of the heart is barely described during AS. The first description was made during World War II, as a result of military operations on the island of Leyte in the Philippines, where more than 1500 American soldiers contracted AS related to *S. japonicum*. An analysis was made of 315 electrocardiograms and found varied repolarisation abnormalities: anomalies of T-waves (99%) or S-T segments (52%). These changes were attributed to the side-effects of antimony, the anti-schistosomiasis drugs used at that moment. According to our knowledge, only one study described cardiac features during an *S. mansoni* epidemic in Brazil. Among the 31 patients who had contracted the disease after bathing in a contaminated lake, 12 people (38.7%) had chest pain and six (19%) had pericarditis diagnosed with ultrasounds. Besides pericarditis, one case of endomyocardial fibrosis has been described in association with central nervous system disorders in a 25-year-old patient infected by *Schistosoma mansoni*. Thus *S. mansoni*, *S. haematobium*, as well as *S. japonicum* have been involved in heart involvement during AS.

Acute myocarditis during AS may be related to the immunological phenomena caused by the presence of schistosomulae in the blood circulation and tissue. Eosinophils play an essential role in the regulation of allergy and in the protective role in the regulation of allergy and in the protec-

**Figure 1.** Electrocardiogram of patient number 1: negative T-Wave in lead VI and depression of the S-T segment in lead V4, V5, V6 and DII.

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