

***Schistosoma mansoni* myelitis in two patients who had traveled to West Africa**

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Schistosomiasis remains a widespread parasitic infection affecting more than 200 million people over three continents. Neurologic involvement, reported at the beginning of the century with *Schistosoma* species, is uncommon but may be severe [1]. Cerebral infection is more likely to be caused by *Schistosoma japonicum*, and spinal cord involvement by both *S. haematobium* and *S. mansoni*. Recently, two patients with spinal cord schistosomiasis due to *S. mansoni* were examined in our department. Magnetic resonance imaging (MRI) proved to be of great value for the diagnosis of schistosomiasis and assessment of the efficacy of therapy in these cases. Moreover, epidemiology linked to these two cases raises speculations concerning the neurotropism of some *S. mansoni* strains.

**Patient 1**

A 21-year-old French patient was admitted to the hospital in August 1993, because of acute urinary retention and weakness of the lower extremities. The patient had traveled to Burkina Faso and stayed there from May 1993 to June 1993. He had followed an appropriate antimalarial prophylactic regimen. During this trip, he swam in a lake near the Banfora district.

On admission, physical examination revealed a painful and distended bladder which was rapidly relieved by a catheter. Neurologic examination showed a slight distal motor deficit with loss of strength in the lower extremity. Deep tendon reflexes were absent at the ankle and left knee, and plantar responses were flexor. Anal sphincter dysfunction was noted, with hypotonia and bowel incontinence. There was some perineal anaesthesia. Backache was noted, involving paraspinal muscles in the lumbar region. Motor and sensory function in the upper extremities and cranial nerves and cerebellar function were normal.

MRI was performed on the same day (Gyroscan Philips 1.5 T and MR max. GE 0.5 T) and revealed moderate widening of the conus medullaris with a central and non-homogeneous T<sub>2</sub> hypersignal in T<sub>2</sub>-weighted sequences as well as heterogeneous enhancement after intravenous injection of gadopentate dimeglumine (0.1 mmol/kg body weight).

Biological tests showed an erythrocyte sedimentation rate of 20 mm/h, and a white blood cell (WBC) count of 11 900/mm<sup>3</sup> with 29% (3451) eosinophils. Coagulation tests and liver function were normal.

Stool examination and rectal biopsy revealed *S. mansoni* eggs. Cerebrospinal fluid (CSF) examination showed a moderate pleiocytosis (60 cells/mm<sup>3</sup>), predominantly lymphocytic but including numerous eosinophils. The CSF protein was 0.95 g/L. The indirect immunofluorescence test for *S. mansoni* antibodies in the serum was positive (1/160) and strongly positive in CSF. Tests for HIV-1, HIV-2 and HTLV-1 viral antibodies and *Borrelia burgdorferi* antibodies, and serologic tests for *Treponema pallidum*, were negative. CSF and blood viral cultures for cytomegalovirus were negative.

Praziquantel (40 mg/kg twice at 12-h intervals) was administered on the fourth day along with methylprednisolone, 10 mg/kg IV, once on the first day and steadily decreasing to a dosage of 1 mg/kg per day orally for 2 weeks. At discharge, on day 20, the patient had improved lower extremity strength, but persistent bladder dysfunction required an indwelling catheter. Four months later the patient was considered cured, with normal neurologic examination and normal sexual and urinary functions. MRI follow-up examination at 2 and 4 months showed normal conus medullaris morphology and signal in both T1 and T2 TSE weighted sequences without enhancing contrast injection.

**Patient 2**

A 22-year-old French patient with no previous history was hospitalized in March 1994 complaining of low back and leg pain together with retention of urine and fecal incontinence. He had traveled to West Africa between February 1993 and August 1993, and had met patient 1 in Burkina Faso in May 1993. He swam in the same lake near the Banfora district. He came back to France in August 1993 and parasitologic investigations were done at this time because of diarrhea. Serology for *S. mansoni*, *S. haematobium* and *S. japonicum* was negative but eosinophilia was noted (3200/mm<sup>3</sup>). No specific treatment was prescribed at that time.

One week before admission, severe low back and leg pain occurred followed by sphincter dysfunction. Only neurologic examination revealed abnormalities. Lower extremity strength was decreased while deep tendon reflexes were normal. There was partial urinary retention and the anal sphincter was hypotonic. Sciatica was noted on both sides. An electromyogram of the lower limb ruled out peripheral neuropathy. MRI of the conus medullaris demonstrated a mild enlargement of the spinal cord with a pseudotumoral appearance in T1- and T2-weighted sequences. Small and heterogeneous areas of intramedullary contrast

uptake were noted after intravenous injection of gadopentate dimeglumine (Figure 1). CSF study demonstrated 59 WBC/mm<sup>3</sup>, mainly lymphocytes, with 13% eosinophils and high proteinorachia (1.10 g/L). No inflammatory syndrome was noted and further biological and serologic tests were normal. Rectal biopsy revealed *S. mansoni* ova.

*S. mansoni* antibodies were present in the patient's serum (indirect immunofluorescence: 1/160) but no antibodies were detected in the CSF. Since all viral and bacterial serologic tests (mainly HIV-1, HIV-2 and HTLV-1) were negative, spinal cord involvement was attributed to a schistosomal infection. The same treatment as given to patient 1 was administered. Rapid improvement of leg strength and sphincter function, and decrease in back and leg pain, were noted. At discharge, on hospital day 15, the patient was walking normally. Two months later, follow-up physical and MRI examination confirmed the recovery without neurologic sequelae.



**Figure 1** Patient 2. Gadolinium-enhanced sagittal T1 weighted MRI scan showing pseudo tumoral aspect of the conus medullaris with intramedullary contrast enhancement after intravenous gadopentate dimeglumine.

### Discussion

Since the adult schistosome does not replicate in its human host, the major factor determining the extent of morbidity is the intensity of infection. Pathogenesis is mainly due to the laying of eggs in tissues, inflammatory reaction, and fibrosis. Eggs can usually be found in the intestine or bladder walls and atypically in many other organs if they fail to reach their normal outlet in the urinary or intestinal tract. Ectopic location of eggs has frequently been reported, and central nervous system (CNS) involvement has been observed [2–4]. The two cases reported here raise interesting diagnostic and epidemiologic questions.

Although less than 100 cases have been reported, four main pathologic forms of spinal cord schistosomiasis have been described: (1) a myelitic form caused by the presence of ova in the spinal cord and the surrounding reaction; (2) compression of the spinal cord by granulomas which may be intra-axial or extra-axial; (3) radicular involvement characterized by the presence of multiple granulomas in spinal roots; and (4) anterior spinal artery occlusion (rare).

MRI findings are non-specific. In our two cases, MRI showed an intramedullary mass suggestive of neoplastic or inflammatory disease, and indicating schistosomal spinal cord involvement in the context of the clinical presentation, epidemiology, serologic tests and CSF study. The regression of MRI signs with specific treatment confirmed the diagnosis. This MRI aspect is the most frequent of those reported. MRI is a very useful tool during neurologic involvement of this parasitic disease, first in suggesting the diagnosis, second in obviating invasive explorations such as myelography or laminectomy for diagnosis, and finally for assessment during follow-up [5].

In both of our cases, stool examination or rectal biopsy detected *S. mansoni* eggs, and specific antibodies were detected in the serum (and in high titers in the first patient's CSF). It has been reported that a higher antibody response in the CNS than in blood may indicate CNS involvement [6].

In 1992, the CDC and the US Peace Corps detected a 33% prevalence of *S. haematobium* infection among expatriates in Malawi. During that year, two US Peace Corps volunteers were evacuated from Africa because of *S. haematobium* infection of the CNS [7]. In 1993, a patient with schistosomiasis of the spinal cord following freshwater exposure in Lake Malawi was also described [8]. Another case of cerebral schistosomiasis caused by *S. haematobium*, probably acquired in Lake Malawi, was reported in 1994 [9]. From the epidemiologic point of view, these four cases of neurologic schistosomiasis and our two cases of spinal cord *S. mansoni* schistosomiasis were all acquired after

swimming in the same type of still waters (Lake Malawi for *S. haematobium* and Burkina Faso for *S. mansoni*). It must be kept in mind that location of the eggs in the spinal cord is due to aberrant migration, so it is rather surprising to note this rare phenomenon in our two patients. It has been previously reported that differences in schistosomiasis-associated morbidity may be observed between areas where prevalence and intensities of infection are comparable [10]. The reasons for these differences are still not clear.

The differences between patients depend on many factors, such as intensity and duration of infection, nutritional state, concomitant infections, differences in pathogenicity between local parasite strains, and host genetic differences [11]. Since the two patients were Caucasian, non-immune and in a normal nutritional state, and since they contracted schistosomiasis under similar conditions, it may be suggested that the spinal cord localization is either a random accident or due to a particular neurotropism of the strain. Intraspecific variation is widespread amongst *Schistosoma* spp. resulting in differences in infection rate, egg distribution in tissues, maturation time, egg-laying capacity, pathogenicity, virulence, and susceptibility to drugs [12,13]. These differences may directly affect the epidemiology of schistosomiasis in various geographic areas. It would be of great interest to analyze the genomic patterns of strains isolated from areas where clinical manifestations are atypical.

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## Acrodermatitis chronica atrophicans and serologic confirmation of infection due to *Borrelia afzelii* and/or *Borrelia garinii* by immunoblot

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Acrodermatitis chronica atrophicans (ACA) is a late chronic cutaneous manifestation of Lyme borreliosis [1], recognized almost exclusively in Europe [2]. Since 1992, European isolates of *Borrelia burgdorferi* sensu lato, the agent causing Lyme disease have been subdivided into three major genospecies: *B. burgdorferi* sensu stricto, *B. garinii* and *B. afzelii* [2,3]. Whereas *B. burgdorferi* sensu stricto seems to be the major if not the only genospecies of clinical importance in North America, European isolates belong to three genospecies, mostly *B. garinii* or *B. afzelii* [4]. Recent findings suggest that these three genospecies are associated with different late clinical manifestations, respectively to Lyme arthritis, neuroborreliosis and ACA [2,5–7].

Because of its sometimes debilitating characteristics and its poor prognosis, ACA should be diagnosed and treated as soon as possible. Although *B. burgdorferi* sensu lato has been successfully cultured from various clinical lesions, culture remains a difficult procedure that is not practicable for routine diagnosis. Clinical diagnosis