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A case of 'tropical' myelopathy

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Abstract

We present a case of lower limb sensory disturbances and weakness in a patient originating from Mali. MRI showed a diffuse myelopathy of the cervical and thoracic spinal cord. Serological evaluation of blood and cerebrospinal fluid pointed towards schistosomiasis as the cause. Histological confirmation was made on bladder-biopsy. Treatment with praziquantel and steroids brought marked clinical improvement. This case illustrates the need to keep in mind more exotic causes of myelopathy in those patients coming from endemic regions.

Key words : Schistosomiasis ; spinal cord ; myelopathy.

Case report

A 43 year-old patient of Malian origin presented to the hospital with complaints of pain and sensory disturbances in both legs. The pain was described as burning and developed progressively, over about 2 years, from the tips of his toes to knee-level. He also complained of weakness of the legs and low back pain. His past medical history was unremarkable except for a gastric ulcer treated by ranitidine. The patient needed fentanyl patches for the invalidating pains.

His last stay in Mali was about three years before presentation, but the patient did not mention swimming outdoor at that time. He had no urinary complaints, and specifically no hematuria. General physical examination was normal. Neurological examination showed hypoesthesia for touch and pinprick as well as diminished vibration sense and proprioception distal in the legs, with flexion and extension paresis of 4-/5. The patient had mild gait instability related to loss of proprioception. MR imaging of the cervico-thoracal spine showed asymmetrical intramedullary hyperintensity on T2 weighted images extending from C1 to Th7 (Fig. 1). Clinical biochemistry and hematology showed no abnormalities. Serological evaluation for HIV, HTLV-1&2, syphilis, hepatitis C and brucellosis was negative. Initial evaluation showed a weak positive result for Borrelia burgdorferi but Western blot was negative. The patient showed

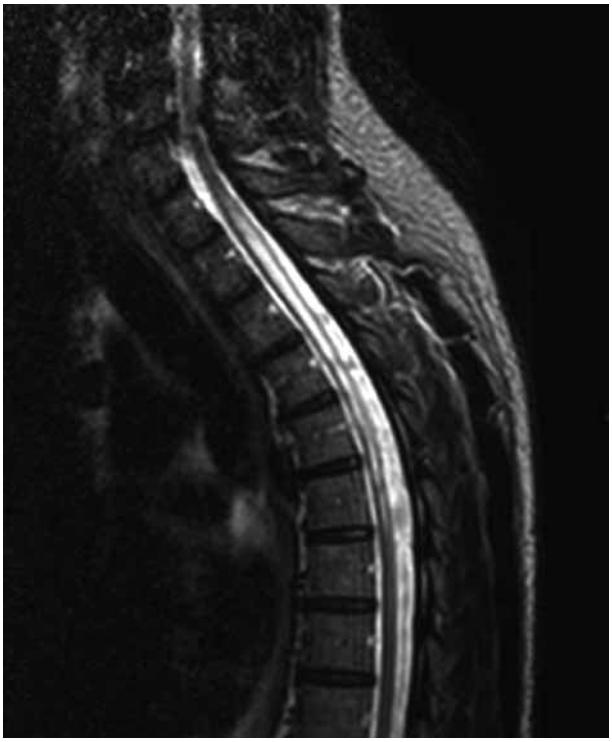

FIG. 1. — T2-weighted image showing an intramedulary hyperintensity suggesting edema extending from C1 to Th7.

serological evidence for a chronic hepatitis B infection. Autoimmune tests were negative except for a 1/80 positive anti-nuclear antigen without specificity. An arteriovenous malformation was ruled out by a spinal angiogram. Serological evaluation for strongyloides, toxocara, ecchinococcus multilocularis/granulosis and taenia were negative ; however, anti-schistosoma antibodies were positive. Urine and faeces samples showed no schistosoma eggs. A CT of the abdomen revealed a diffusely thickened bladder wall with nodular components (Fig. 2). Cerebro-spinal fluid (CSF) analysis was strictly normal but serological tests on CSF confirmed intrathecal synthesis of schistosoma antibodies. A biopsy of the bladder wall showed several schistosoma eggs (Fig. 3). Treatment with praziquantel at 60 mg/kg/day for 3 days was started together with methylprednisolone at 64 mg/day.

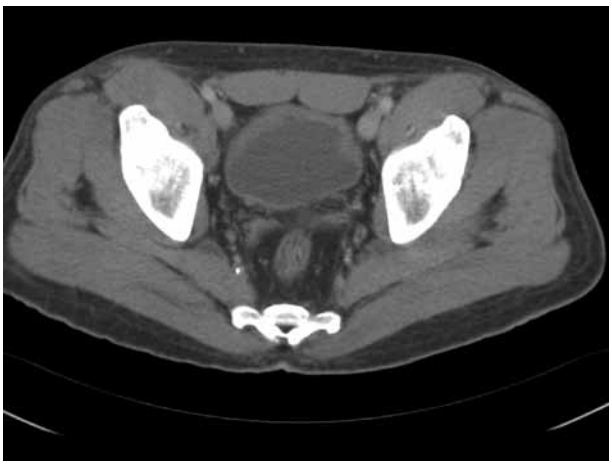

FIG. 2. — CT of the abdomen showing diffuse thickening of the bladder wall.

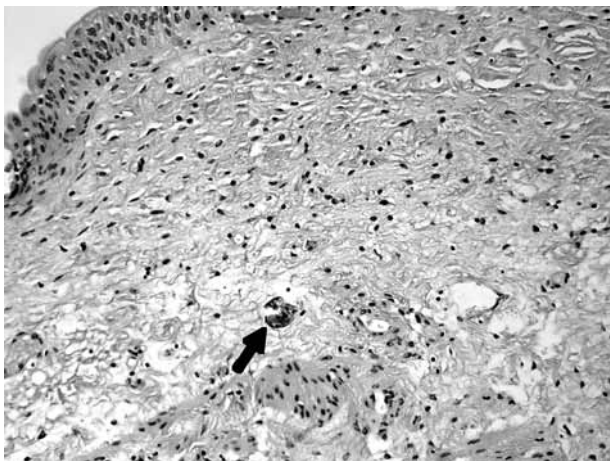

FIG. 3. — Bladder biopsy showing normal urothelium, sclerotic thickening of the propria, minimal inflammatory reaction with a few eosinophils and, almost in the muscularis mucosae, a schistosome egg (arrow).

In the weeks following treatment the patient showed resolution of the pain and sensitive symptoms, as well as marked strength improvement.

Discussion

In 1851 Theodor Maximilian Bilharz identified the schistosoma trematode as the causative agent of the so-called "tropical hematuria" (1). Schistosomisiasis or "bilharziosis" is caused by a blood fluke and is the second leading parasitic disease after malaria, with about 200 million people affected worldwide (2). Moreover in Mali (population : about 12 million), approximately 2.5 million persons have urinary tract schistosomiasis with S. haematobium (3). Infection is transmitted by specific snails whose distribution determines the prevalence of the three different species of schistosoma responsible for most disease in humans : S. haematobium (frequent in Africa, affecting the bladder and urinary system, rarely spinal cord/roots and very rarely the brain), S. mansoni (common in the Caribean, South America and Middle East besides Africa, mainly causing infection of the liver and rectum, rarely spinal cord/roots and very rarely the brain), and S. japonicum (frequent in Japan, China and South-east Asia, affecting intestines and liver, rarely the brain) (4). In 1905, Shimamura and Tsunoda demonstrated the first case of transverse myelitis due to schistosomiasis (5). Myelopathy is the consequence of ectopic schistosome ova deposition with immune-mediated tissue damage and granuloma formation (4). Although schistosomiasis most frequently attacks the lower lumbar spine and cauda equina, cases have been published with isolated cervical lesions (6, 7). The clinical manifestation is most frequently low back pain followed by weakness and sensory loss in the lower limbs associated with bladder dysfunction (4). MRI of the spine usually shows hyperintense T2-weighted lesions, that are iso-intense on T1 images. Contrast enhancement has frequently been described and a reactive syrinx can be present (8). CSF changes in spinal schistosomiasis have been described, characteristic features are mild to moderate pleocytosis with presence of eosinophils, slight to moderate protein increase, elevated gamma globulin concentration and a positive immune assay. A series of 22 patients (9) however showed CSF eosinophilia in only 33,8% of patients, with 13.7% of the patients having normal CSF-tests. Testing for antibodies was suggested to be the most specific test for the diagnosis of spinal schistosomiasis. Treatment of spinal schistosomiasis consists of praziquantel 60 mg/kg/day for 3 days with prednisolone 1.5 to 2 mg/kg for three weeks, gradually tapered thereafter (4).

In summary, spinal schistosomiasis should be considered in the differential diagnosis of unexplained myelopathy in patients coming from endemic regions. The present report illustrates the need for extensive check-up to avoid unnecessary spinal cord biopsy.

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