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Systemic schistosmiasis with longitudinal extensive transverse myelitis (LETM) and mild optic neuritis: Case report

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Abstract

Spinal cord schistosomiasis is difficult to diagnose in nonendemic areas. We report the clinical profile of young Saudi male who presented with myelopathy and optic neuritis. He had received both pulse steroid therapy and a plasma exchange. Praziquantel was administered late and the patient did not recover. We concluded that prompt recognition and early treatment with praziquantel is crucial for a better outcome. The role of steroids in these cases still needs to be proven.

Keywords

neuroschistosomiasis; optic neuritis

Introduction

Schistosomiasis is a parasitic infection caused by different species of schistosomal parasites. Schistosoma haematobium (endemic in Africa and Mediterranean countries), *S. mansoni* (endemic in Africa and the Middle East) and *S. japonicum* (endemic in Japan and China). Data reported in 2016 from 36 countries for treatment of school-aged children and from 21 countries for treatment of adults show that more than 86 million people received preventive chemotherapy for schistosomiasis globally, which is equivalent to 52% global coverage for school-aged children and 14% for adults [1]. The infection occurs following contact with fresh water containing snails [2]. Schistosomal infection can affect any part of the body. The incubation period usually lasts from weeks to months [3]. Neuroschistosomiasis refers to a schistosomal infection of the brain or spinal cord, it may cause disabling disease if not treated promptly [3]. There were 2 studies published about schistosomal infection with optic neuritis, first study was to describe the demographic, clinical and laboratory features of Idiopathic Acute Transverse Myelitis (IATM), 70 of the 70 cases of acute myelopathies, the idiopathic form was identified in 41 following exclusion of the cases associated with systemic lupus erythematosus (n=1), Sjogren's syndrome (n=1), herpes zoster (n=1), cytomegalovirus in an HIV-positive patient (n=1), Schistosoma mansoni (n=1), actinic myelitis (n=1), infectious myelitis of unknown etiology (n=2) and those that, following the first attack of myelitis [6]. Second case report a

woman presenting with cough, fatigue, atypical optic neuritis with chiasmitis. A previous exposure history; serological testing confirmed schistosoma infection, treated with praziquantel and slowly improved clinically [7].

In Saudi Arabia we found only 2 cases were reported none of them was associated with optic neuritis, first one was produced at 1994 from King Faisal Specialist Hospital & Research Center in Riyadh, a case of intramedullary neuroschistosomal granuloma in a 15-year-old girl is reported [8]. Second case was from National Guard; they report the clinical profile of 2 young Saudi males who presented with myelopathy. The first patient arrived at our hospital relatively late, 3 months following the presentation of initial symptoms, and had received both pulse steroid therapy and a plasma exchange. Praziquantel was administered late and the patient did not recover. The second case presented early, within around 8 weeks of initial symptoms. This patient received praziquantel without any kind of steroid and had a complete recovery [9].

We are reporting a case of myelopathy caused by schistosomal infection with rare neurological manifestations in form of optic neuritis.

Aim: To report and describe a very rare clinical presentation of Schistosomiasis, which is Optic neuritis and Extensive transverse myelitis.

The Case

He is a 14-year-old Saudi boy, medically free, from Riyadh who presented with 3-day history of acute-onset, progressive-course severe lower back pain, followed by progressive lower limb weak-ness, urinary retention. There was history of upper respiratory tract infection symptoms (cough, runny nose) and fever lasting for about 10 days, 23 weeks prior to presentation. The patient gave a remote history of swimming in contaminated wells since long time. Examination unremarkable except lower limbs weakness in his legs, which was more pronounced proximally and worse in the left leg. Deep tendon reflexes were +1 symmetrical and there was no clear sensory level, however the patient had hypersensitivity in his legs bilaterally.

The initial MRI showed a long segment of central abnormal signal intensity in T2 to superior endplate of T8 to the conus medullaris as shown in figure 1. Brain MRI showed faint enhancement going with mild bilateral optic neuritis shown below in figure 2. Cerebrospinal fluid (CSF) analysis showed high protein 1.07 and WBC 43 and he was negative for infection and cytology. His Schistosoma titer was negative at a ratio of 1:80. Visual evoked potential showed P100 latency of 125ms in the right eye and P100 latency of 126ms in the left. CT chest, abdomen and pelvis showed bilateral ill-defined multiple lung nodules, multiple perirectal mildly enlarged lymph nodes in the mesorectal fascia, smaller presacral lymph nodes and liver cyst. Colonoscopy with biopsy from terminal ileum showed focal granuloma with an oval foreign body highly suspicious for Schistosoma egg. Repeated serum Schistosomal antibody titer was high this time (1:640). The patient had been treated earlier (before the biopsy) as acute transverse myelitis with 7days course of pulse steroid with mild clinical improvement only, and had been enrolled in an extensive rehabilitation program. The patient had received praziquantel and continued on steroids. His rehabilitation was

without significant improvement. Follow up MRI spine showed improvement in terms of decreased enhancement and swelling of the conus medullaris. He stayed in the hospital for around three months. He was later and was found to have mild improvement, but he remained wheelchair bound. He was able to make minimal movements with both lower limbs but not against gravity. The patient exhibited urinary retention using catheter, yet he had no erectile dysfunction.

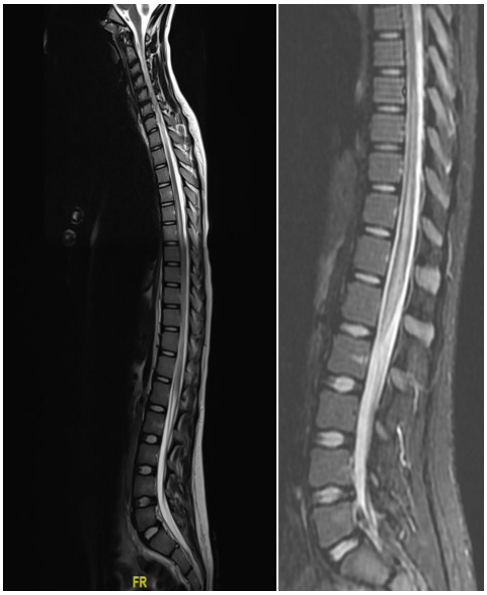

Figure 1: There is a long segment of central abnormal signal intensity in T2 to superior endplate of T8 to the conus medullaris.

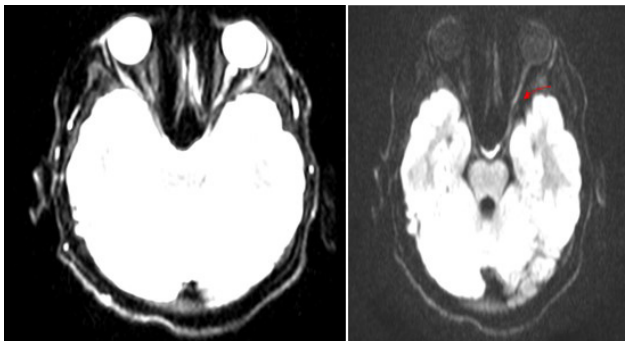

Figure 2: MRI brain with contrast (T1) showing contrast enhancement of left optic nerve indicating left optic neuritis.

Discussion

Neuroschistosomiasis is considered disease due to the fact that it is not usually symptomatic or may be difficult to diagnose based on the clinical background or serological testing [4]. Therefore, high suspicion of myeloschistosomiasis should be attained during the assessment of nontraumatic back pain of ypung patients presenting with transverse myelitis symptoms and signs, particularly if there is a history of traveling to endemic areas [2]. Status of schistosomiasis not yet determined in Saudi Arabia [1]. Myeloschistosomiasis has a predilection for the lower spine, involving especially the lower thoracic (T6), like in our case, to the upper lumber spinal cord segments. It is generally associated with sensory level, bladder, and bowel dysfunction [4]. The amount of time between the beginning of the symptoms and the establishment of the complete manifestation normally ranges from a few days to a couple of weeks [4]. Neuroschistosomiasis can present up to 3 years after exposure [5]. The patient did swim in a contaminated well long time before

the start of the symptoms. We believe that this could be the source of infection. Rectal biopsy can detect the Schistosoma eggs only in 42% of cases, as in our case, [2]. A CSF analysis will usually show pleocytosis and high protein content [2]. Another diagnostic modality is an MRI, which usually shows intramedullary expansion signals. Treatment for myeloschistosomiasis includes the anthelmintic praziquantel. It is recommended to be preceded by a steroid therapy in order to avoid the immune response that usually occurs during starting praziquantel [2–4]. Most reported cases showed improvement after commencing therapy promptly [4]. In conclusion, spinal cord schistosomiasis can cause significant disability if not treated promptly with praziquantel or, especially, if treated with steroids alone in the beginning. Although the use of steroids is highly recommended in the literature. The use of steroids is still unproven and will be better judged by well-constructed randomized controlled trials.

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