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Case Report

Arborized pattern of MRI enhancement in spinal cord schistosomiasis: A report of 2 successful case outcomes ☆

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ABSTRACT

This case report describes 2 patients with spinal cord schistosomiasis diagnosed based on a magnetic resonance imaging finding of a unique arborized type of postcontrast enhancement. Both patients presented with back pain and lower limb weakness, and prompt treatment with an anti-schistomal agent and steroid resulted in significant neurological and radiological improvement. The report emphasizes the role of imaging in the early diagnosis of spinal cord schistosomiasis, as well as the importance of early treatment for the best clinical outcomes.

Introduction

Schistosomiasis is a parasitic blood fluke (trematode) infection spread by contact with fresh water sources contaminated by Schistosoma harboring snails, which are the intermediate hosts for transmission [1,2]. The 3 commonest species are *S. haematobium*, *S. mansoni*, and *S. japonicum*. It is a neglected tropical disease with the highest disease burden seen in sub-Saharan Africa [3–6]. Once infection establishes, the location of egg deposition in the definitive human host determines the affected systems [1,2]. Neuroschistosomiasis is an uncommon but severe form of the disease affecting the brain and spinal cord [7–11]. Eggs in the brain and spinal cord cause a wide range of damage by triggering a granulomatous inflammatory response [1,2,7]. Spinal cord schistosomiasis (SCS) can cause acute to subacute back pain, motor weakness and paresthesia in the lower limbs, autonomic dysfunction, and transverse myelitis syndrome [8,9]. While histopathology provides a definitive diagnosis, its invasive nature discourages the collection of surgical samples [10]. A presumptive diagnosis is based on history, physical examination, supportive labora-

Abbreviations: SCS, spinal cord schistosomiasis; MRI, magnetic resonance imaging.

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tory examinations, and imaging findings [9,10]. Magnetic resonance imaging (MRI) is the imaging modality of choice, and it commonly shows lumbosacral T2 hyperintensities, enlarged cord and cauda equine roots, and heterogeneous enhancement [2,9,11]. A new MRI finding on the T1W postcontrast sequence is thought to be unique; it shows an arborized pattern of enhancement with a central linear region and punctate nodules around it [7]. Early treatment initiation is required for the best treatment outcomes, and it has resulted in complete clinical and radiological resolution [7,12,13]. Praziquantel, the cornerstone of schistosomiasis treatment, is an effective agent for all species of schistosoma [14,15], and although no controlled studies exist, steroids are recommended to limit the mass effect and an acute allergic reaction related to Praziquantel treatment in SCS [2,9,10]. The role of surgical therapy is controversial, but it is required in cases of severe neurological complications and to obtain a pathological sample [8,16].

Case presentations

Case 1

Our first patient is a 12-year-old male who presented with an acute onset of lower back pain and bilateral leg weakness lasting 2 weeks. He had no sensory symptoms, complaints of urinary retention, or fecal incontinence. Prior to this, he was in good health, had normal growth, and had never required hospitalization. On multiple occasions, he provided a positive history of fresh water exposure near his residence. On physical examination, he had normal vital signs, including a normal temperature of 36.6 degrees Celsius. The neurologic examination revealed normal and comparable muscle bulk in his thigh and leg muscles, as well as decreased power bilaterally (3/5). He also had knee hyperreflexia. His basic investigations, including complete blood count, erythrocyte sedimentation rate, and organ function tests, were unremarkable. We subsequently performed a lumbosacral MRI (Figs. 1A-C). It showed an edematous cord at the conus medullaris level with a linear and nodular enhancement pattern, giving the arborized enhancement pattern. The imaging pattern was highly supportive of spinal schistosomiasis. Following the MRI findings, a Kato Kanz microscopic examination of his thick fecal smear yielded positive *S. mansoni* eggs. The suggestive imaging findings and positive stool findings led to treatment initiation with praziquantel 60 mg/kg/day in divided doses for 5 days. Follow-up MRI (1D-F), 5 months from the treatment period, was remarkable for markedly decreased edema and enhancing regions.

Case 2

The second patient, a 14-year-old male, came with the chief complaint of gradually progressing weakness in both his legs for 40 days. The weakness was worse on the right side, and at presentation, the patient had lower back pain, which further contributed to difficult ambulation. He did not complain of any sensory, bowel, or bladder problems. The patient was a previously healthy and thriving student with no chronic illnesses. He lives in a schistosomiasis-endemic area and frequently swims in a lake. On examination, his vital signs were normal. On a neurologic examination, power over the right thigh and leg was 3/5, and on the left, 4/5. He had a right knee clonus, and his right side increased his plantar reflex. The patient, residing in an endemic region, underwent a urine and stool Kato Kanz test. His stool showed *S. mansoni* eggs. Other relevant laboratory examinations, including a complete blood test (White Blood Cells = 8500 cells/microliter with a Polymorphonuclear differential of 60%), erythrocyte sedimentation rate (12 mm/hour), and organ function tests, were normal. Cerebrospinal fluid analysis showed a non-specific rise in mononuclear proportion. A spinal MRI (Figs. 2A–C) was performed for further assessment. There were linear, nodular, central, and peripheral enhancing regions in the edematous conus medullaris that were more conspicuous on the postcontrast sequences. Consequently, a decision was made to treat the patient with divided doses of Praziquantel (60 mg/kg/day) for 5 days and parentral methylprednisolone with tapered doses of oral prednisolone. The patient made good clinical progress, and his right leg power improved to 4/5. A 2-month follow-up MRI (Figs. 2D-F) depicted reduced cord edema as well as enhanced regions.

Discussion

Schistosomiasis affects an estimated 250 million people worldwide, and 90% of those requiring treatment live in Africa [3,4]. Ethiopia is one of the most endemic countries for schistosomiasis, with about 5 million people affected [5,6]. Neuroschistosomiasis is uncommon, and only 800 cases have been reported since its first description in 1930 [9]. *S. mansoni* primarily is the main cause, with egg deposition in the spinal cord ranging from 0.3% to 13% [17]. Without prompt treatment, the prognosis is poor [2,9,11]. Young males are more susceptible as a result of their greater occupational exposure to contaminated water sources [2,10,16,17]. Saleem et al. [11] reported SCS in 8 male patients, whose mean age was 16.7 years.

Two theories explain how schistosoma eggs reach and deposit in the spinal cord: the first theory hypothesizes that eggs travel hematogenously with subsequent egg deposition anywhere along the spinal cord's length, and the second theory postulates in situ deposition of eggs by adult worms after 'ectopic' travel through leptomeningeal veins during the early stages of infection [7,10,18]. The disease has acute-to-subacute, usually radicular, and mild-to-severe lower back pain. Lower limb weakness, paresthesia, associated bladder and bowel dysfunction, and deep tendon reflexes subsequently follow. Neglecting lower back pain as a serious symptom, as well as the similarity of the condition to neoplastic diseases, can lead to a delayed diagnosis [9].

A definitive diagnosis of SCS necessitates a histopathological examination of biopsy samples [10]. However, this is not advised as it can cause additional injury to already damaged spinal cord tissue. Therefore, a presumptive diagnosis is

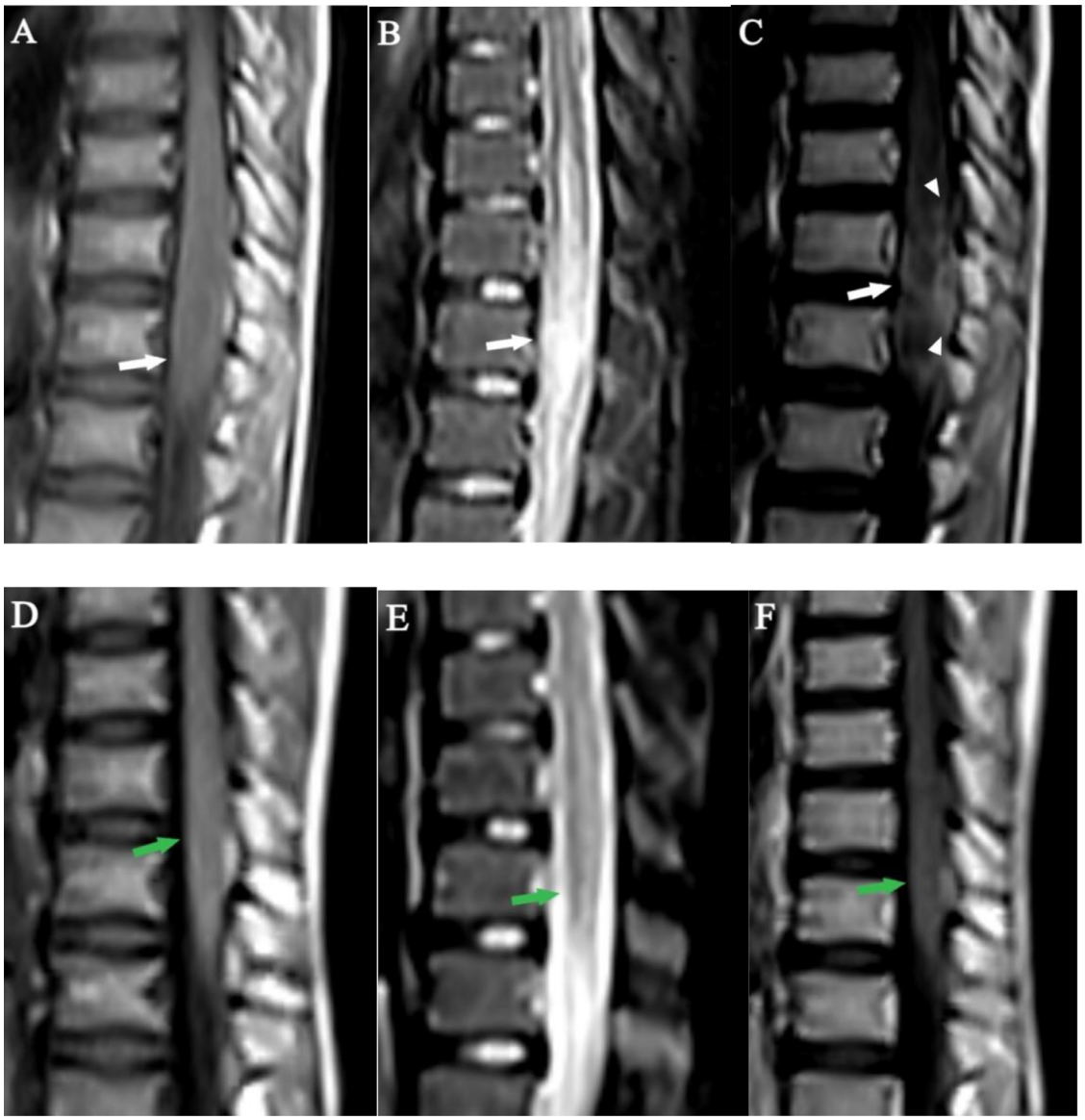

Fig. 1 – Sagittal lumbosacral MRI at initial presentation in T1W (A), T2W (B), and T1 C+ (C) sequences show conal expansion with high T2 signal and surrounding edema (white arrows in A and B). The post-contrast image shows the expanded cord (C) with a peripheral linear-like and nodular type of enhancement giving the arborized appearance of enhancement. On the 5 month post-treatment follow-up MRI in T1W (D), T2W (E), and T1 +C (F) sequences, significant reduction in the expansion of the conus and enhancement were seen (green arrows on D-F).

based on clinical, laboratory, and imaging studies [10]. Urine and stool ova tests are also supportive diagnostic methods [2,10]. CSF examination can provide a nonspecific pattern with raised protein and mononuclear cells [9,10].

MRI is a sensitive tool for diagnosing and managing SCS. It can reveal the true extent of the disease and suggest a diagnosis through recognition of its signal intensity and enhancement changes [11]. An MRI scan will often show T2 hyperintensities in the lower back and neck, spinal cord enlargement, especially in the lower cord, thickening of the cauda equina roots, and various T1 enhancement patterns. Computed tomography is less sensitive and shows cord enlargement with a nonspecific enhancement pattern [8,9,11].

In 2001, Sanelli et al. [7] reported a new MRI ''arborized'' appearance with a linear and nodular enhancement pattern in 3 patients with confirmed neuroschistosomiasis and suggested this pattern as a specific sign of neuroschistosomiasis. Two of the patients had cerebral schistosomiasis, and one had spinal involvement. In the latter case, MRI revealed diffuse T2 hyperintensity with expansion of the distal spinal cord and conus medullaris. The contrast-enhanced T1W images showed a central linear enhancement surrounded by punctate nodules. The brain cases were located in the left temporal and right parietal lobes. T2 images showed vasogenic edema with a mass effect. On contrast-enhanced T1W images, multiple punctate (1-2 mm) nodules surround the cen-

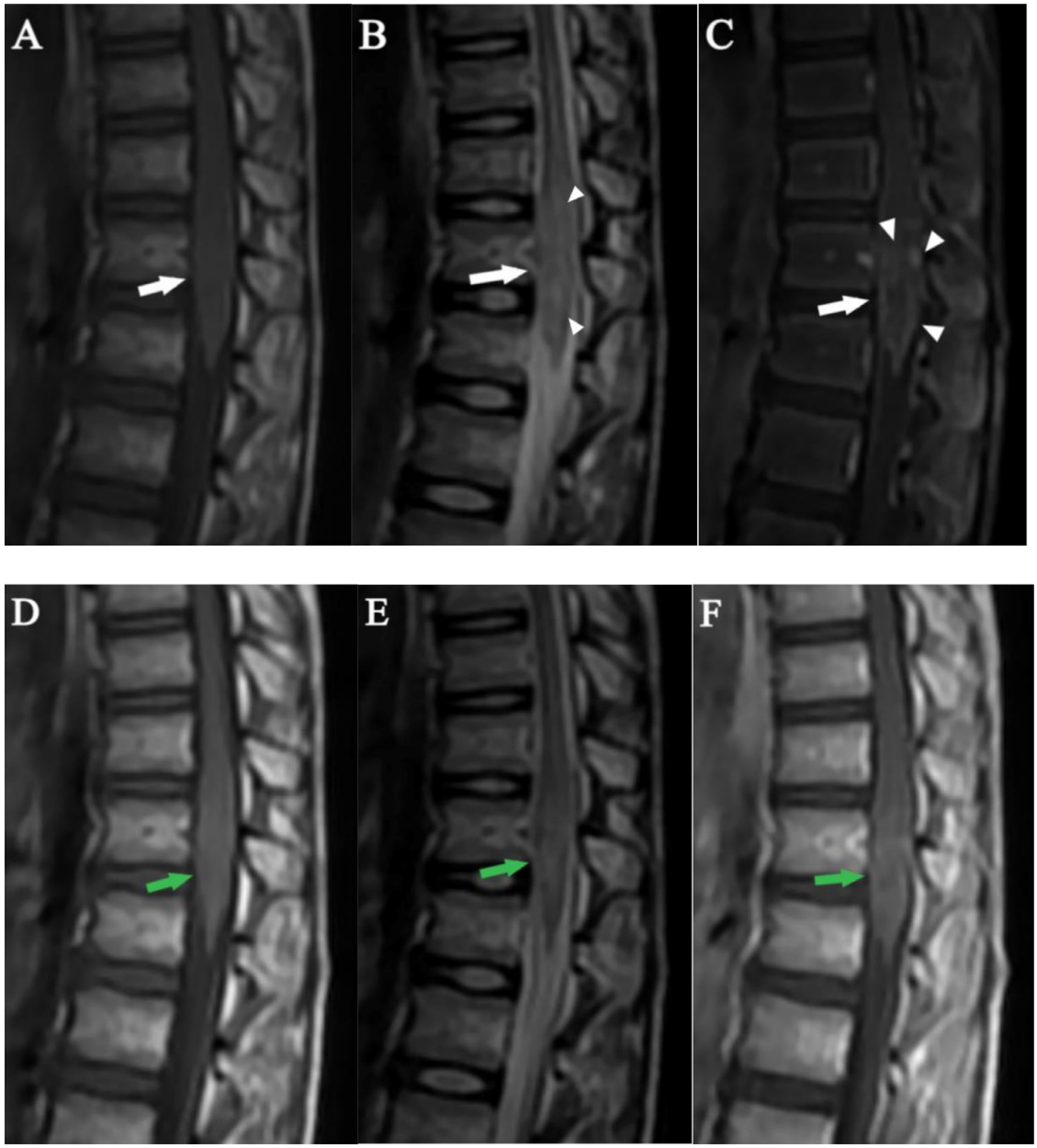

Fig. 2 – Pretreatment sagittal T1W (A), T2W (B), and T1 C+ (C) lumbosacral MRI images show expansion of the conus (white arrows in A-C). On the T2W image, there was a central linear hyperintense signal (white arrowheads on B). Furthermore, peripheral nodular-linear pattern of enhancement was surrounding this central signal on the post contrast image (white arrowheads on C). On the 2 month post-treatment follow-up MRI in T1W (D), T2W (E), and T1 +C (F) sequences, there was a reduction in the level of the conus swelling and marked decrement in enhancement (green arrows in D-F). There was still some central and peripheral residual enhancement seen.

tral linear enhancement. In all 3 cases, the enhancement resulted in a clustered appearance with an arborized pattern. The histopathologic correlation revealed that the granulomas forming around the enhancing punctate nodules cause swelling and blood vessel blockage. We thought that the slow blood flow in the central linear-enhancing region was a result of adult worm migration to the leptomeningeal veins.

Nonrecognition of SCS on imaging can be secondary to its rarity and confusion with other common conditions. One case report from Ethiopia [19] reported a 4-year-old child who presented with complete paraplegia of the bilateral lower limb. The initial MRI was read as an astrocytoma. A biopsy at a later point revealed Schistosoma parasites with extensive necrosis and inflammation. His severe neurologic deficits persisted despite medical treatment, compounded by a pressure abscess of the gluteal region. Upon closer examination of the provided sagittal postcontrast MRI images, we observed a discontinuous linear enhancing region, encircled by prominent, closely approximated nodules, which are typical of the arborizing sign of SCS. Another report [12] also thoroughly

documents the unfavorable results of delayed treatment initiation.

A dural arteriovenous fistula is an important differential diagnosis for lower thoracic levels and conus medullaris edema that produces longitudinally oriented or multi-segmental intramedullary T2 hyperintensity and T1 hypointensity [20]. More specific findings include serpentine dark flow voids on the ventral and dorsal cord surfaces that span at least 3 spinal levels and show vascular enhancement [20,21]. The condition is more common in the sixth decade of life, with 63.5 years as the mean age in 1 retrospective study [21] . The age of our 2 patients, together with the absence of serpentine dural surface vessels on both initial and follow-up MRIs, made us less persuasive about the diagnosis.

Praziquantel is the drug of choice for schistosomiasis and is active against all species of Schistosoma, with a cure rate of 70%–90% [8]. Steroid augmentation is also recommended against acute allergic reactions (that can follow praziquantel initiation) and the mass effect caused by the granulomatous inflammation [2,9]. MRI changes are not immediate and occur months after treatment initiation [7,9]. Patients who present with severe compression symptoms or who fail medical treatment should consider laminectomy for spinal cord decompression [8]. Resection of the granulomatous lesion is controversial with variable advice, as some think it does more damage and others insist on its effectiveness [8,11,13,16].

In conclusion, the 2 patients in this report had SCS-specific MRI appearances. The unique arborized enhancement pattern seen in both patients was a useful diagnostic clue for spinal cord schistosomiasis, making it easier to tell it apart from other spinal cord diseases. The follow-up MRI, which shows the disease's significant improvement after medical treatment, further supports the significance of this unique enhancement pattern as a diagnostic indicator. Early recognition of the sign can lead to a prompt, noninvasive diagnosis of SCS and the initiation of appropriate treatment, ultimately contributing to improved patient outcomes.

Patient consent

Written informed consent was obtained from the parents of the two patients to publish this case report. Personal identifiers are not used in this paper.

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