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# Case Report



# Spinal cord Schistosomiasis: A child's case with an unsatisfactory outcome that mimicked an intramedullary neoplasm. A rare case report

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#### ABSTRACT

Neuroshcistosomiasis is a life-threatening complication of schistosomiasis. Its prevalence in endemic populations is believed to be underreported at 1–4%. We report a four-year-old child who came to our hospital relatively late since the presentation of weakness and incontinence. MRI revealed long segment (T12-L1) ill-defined intramedullary lesions with cord extension having patchy enhancements. Despite the administration of both praziquantel and pulse prednisolone treatment, there was no convincing result. We thus question the role of steroids and praziquantel in individuals with late-diagnosed spinal schistosomiasis.

# 1. Introduction

Neuroshcistosomiasis is a life-threatening complication of schistosomiasis. Its prevalence in endemic populations is believed to be underreported at 1–4%. Schistosomiasis is a disease caused by parasitic blood flukes. It is acquired through contact with contaminated freshwater [1]

More than two million people are affected worldwide, with considerable morbidity and mortality. The highest burden of infection lies within sub-Saharan Africa, although there is a significant burden of disease in other regions of Africa, the Middle East, Asia, and South America [2–5].

NeuroSchistosomiasis is a rare presentation and can cause severe neurologic complications, including the spinal cord and brain. There are few case reports regarding NeuroSchistosomiasis reported in the literature. Here, we report a case of a four-year-old child who came with delayed presentation of compressive myelopathy.

# 1.1. Case report

A 4-year-old preschool child presented lower extremity weakness of 1-month duration. Initially starting from the right and then involving both legs the next day. Associated with this, he has urinary and fecal incontinence at the same time. Prior to the lower extremity weakness, there was a preceding abdominal pain. He later developed urinary

retention, for which he was catheterized in a nearby General hospital, where he was referred to our hospital for further workup.

At the presentation in the ER, he was conscious with vital signs of PR: 123 RR: 40 T: 37.2 SPO2: 98%, normal anthropometry. Pertinent neurologic finding: hypotonic lower limb with Power of 0/5, Reflexes: 1/5 on patellar and ankle reflexes, loss of sensation in lower limbs till mid umbilicus. Initial investigation revealed CBC: [WBC: 8300 N: 47.6% L: 42% Hg: 14.8 g/dl Hct: 43.8% Mcv: 76.3Fl Mchc: 33.8 g/dl Plt: 437,000], negative stool exam. MRI showed long segment (T12-L1) ill-defined intramedullary T1 hypo intense (Fig. 1) and T2 hyperintense lesions (Fig. 2) with cord extension having patchy enhancements. The post-contrast sagittal image showed intramedullary enhancing lesions (Fig. 3)

While entertaining with the working diagnosis of? "Spinal Astrocytoma," the patient disappeared from the emergency to go to a holly place. Unfortunately, there was no improvement in the patient's condition, and he returned to the hospital after one month. At this time, he was scheduled for surgery, laminectomy was done, and the biopsy was subjected to pathology, which displayed schistosome parasites having a ventral horn (Fig. 4) and also extensive areas of necrosis surrounding inflammatory infiltrates (Fig. 5).

After the biopsy result, the patient started five days of praziquantel (50mg/kg daily) and prednisolone (2mg/kg/day) with anti-acid syrup and was discharged home. Follow-up pediatric neurology clinic after three weeks revealed flickering of toes with no other neurologic

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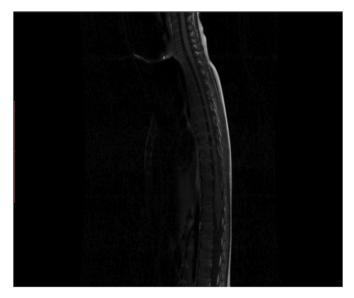


Fig. 1. long segment (T12-L1) MR with ill-defined intramedullary T1 hypo intense lesions.

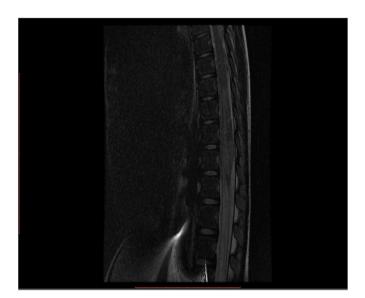


Fig. 2. long segment (T12-L1) MR with ill-defined intramedullary T2 hyper intense lesions.

improvement. He started physiotherapy, and low-dose steroid tapering continued.

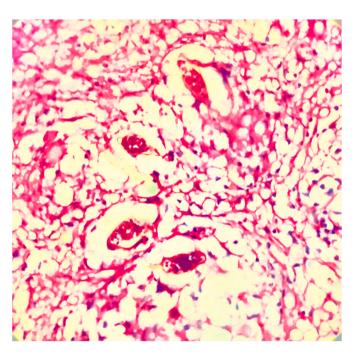
On his 2nd follow-up after a month, he came with similar neurological findings with a left gluteal 5\*6cm indurated tender mass with discharge (Gluteal abscess) due to a prolonged bedridden state for which incision and drainage were done and discharged with oral antibiotics. In subsequent phone call follow-ups, the family denied a rewarding improvement regarding the patient's neurologic illness. This work has been reported in line with the SCARE 2020 criteria [6].

# 2. Discussion

NeuroSchistosomiasis is a rare presentation and can cause severe neurologic complications, including the spinal cord and/or brain. Spinal schistosomiasis is estimated to account for 1–4% and is probably underestimated in endemic populations [7]. It is assumed that embolization of the adult worms to the cord or cerebral microcirculation with subsequent release of eggs leads to an intense inflammatory



Fig. 3. Post contrast sagittal image with intramedullary enhancing lesions.

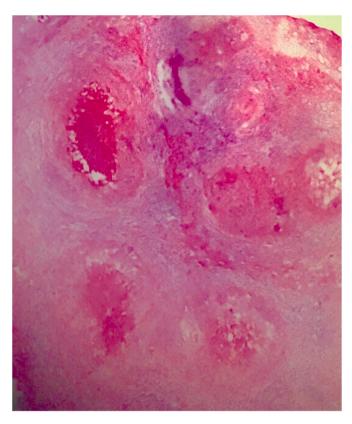


**Fig. 4.** high power view microscope showing four schistosome eggs having a ventral horn.

granulomatous reaction with local tissue destruction and scaring [8]. Mature schistosomes are blood-dwelling trematode helminths that expel eggs via the urine (S. haematobium) and faeces (S. mansoni, S. japonicum). Eggs that are not excreted can become trapped in human tissues causing a peri-ovular granulomatous inflammatory reaction which is the primary mechanism of disease in schistosomiasis [1].

Diagnosis is challenging in non-endemic areas, and travel history must be included in such cases. In this case, despite clinical presentation and the endemicity of the disease, there was no history of river water contact. Most patients present lower back pain, motor deficit, sensory disturbance, and double incontinence [1,9].

A complete blood count may reveal eosinophilia. Stool examination shows eggs nearly sixty percent [10]. Another diagnostic modality is an MRI scan which usually shows intramedullary expansion signals. Ashour et al. reported intramedullary enhancing lesion, this is consistent with our case. The definitive diagnosis necessitates a spinal cord biopsy to



**Fig. 5.** low power view displaying extensive areas of necrosis surrounding inflammatory infiltrates.

demonstrate egg granuloma in the specimen [11]. Despite the administration of both praziquantel and pulse prednisolone treatment precede with laminectomy, there was no convincing outcome. Ferrari TC et al. and his collogues reported a large cohort of 63 cases with absence recovery rate of 14% after administering steroid and anti-parasite.

# 3. Conclusion

In summary, intramedullary signal expansions are regularly seen on MRI, they are not exclusively associated with spinal schistosomiasis. Spinal cord schistosomiasis is an unusual presentation even in endemic areas. There should be a high index of suspicion in patients exhibiting acute compressive myelopathy manifestations.

# Ethical approval

Since it was a case report there were no required approval paper from the Institutional Review Board(IRB)

# Please state any sources of funding for your research

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# Please state any conflicts of interest

The authors declare no conflict of interest.

#### **Author contribution**

All authors contributed in this case report.

# Registration of research studies

Name of the registry:

Unique Identifying number or registration ID:

Hyperlink to your specific registration (must be publicly accessible and will be checked):

#### Consent

Written informed consent was obtained and signed for the parents for the publication of this case report.

# Guarantor

I accept full responsibility for the work of this study.

# Provenance and peer review

Not commissioned, externally peer reviewed.

#### References

- T.C. Ferrari, P.R. Moreira, Neuroschistosomiasis: clinical symptoms and pathogenesis, Lancet Neurol. 10 (9) (2011 Sep) 853–864.
- [2] C.H. King, K. Dickman, D.J. Tisch, Reassessment of the cost of chronic helmintic infection: a meta-analysis of disability-related outcomes in endemic schistosomiasis, Lancet 365 (9470) (2005) 1561.
- [3] J.A. Ultroska, M.G. Chen, H. Dixon, et al., An estimate of global needs for praziquantel within schistosomiasis control programmes, in: WHO/SCHISTO/ 89.102. Division of Control of Tropical Diseases, WHO, Gevena, 1989.
- [4] L. Chitsulo, D. Engels, A. Montresor, L. Savioli, The global status of schistosomiasis and its control, Acta Trop. 77 (1) (2000) 41.
- [5] C.D. Mathers, M. Ezzati, A.D. Lopez, Measuring the burden of neglected tropical diseases: the global burden of disease framework, PLoS Neglected Trop. Dis. 1 (2) (2007) e114. Epub 2007 Nov 7.
- [6] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, for the SCARE Group, The SCARE 2020 guideline: updating consensus surgical CAse REport (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230.
- [7] C.W. Naus, J. Chipwete, L.G. Visser, E.E. Zijlstra, L. van Lieshout, The contribution made by Schistosoma infection to non-traumatic disorders of the spinal cord in Malawi, Ann. Trop. Med. Parasitol. 97 (7) (2003) 711.
- [8] Involvement of the central nervous system in Schistosoma mansoni and S. haematobium infection, A review. Scrimgeour EM, Gajdusek DC, Brain. 108 (Pt 4) (1985) 1023.
- [9] Vale TC, de Sousa-Pereira SR, Ribas JG, Lambertucci JR. Neuroschistosomiasis Mansoni: Literature Review and Guidelines.
- [10] F.J. Carod-Artal, Neurological complications of Schistosoma infection, Trans. R. Soc. Trop. Med. Hyg. 102 (2008) 107–116.
- [11] T.C. Ferrari, P.R. Moreira, A.S. Cunha, Clinical characterization of neuroschistosomiasis due to Schistosoma mansoni and its treatment, Acta Trop. 108 (2008) 89–97.