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Case Report

Intramedullary spinal schistosomiasis in a child with acute myelopathy: A case report

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ABSTRACT

Background: Neuroschistosomiasis is defined as an infection of the nervous system caused by *Schistosoma mansoni*. Neuroschistosomiasis is an important differential diagnostic consideration in pediatric patients presenting with myelopathy. Surgical excision combined with antiparasitic drugs typically provides a satisfactory outcome and often results in neurological recovery.

Case Description: A 4-year-old child presented with acute and progressive myelopathy. A thoracolumbar magnetic resonance image revealed a T12-L2 conus medullaris mass that was isointense on T1 and hyperintense on T2 (with an extensive syringomyelia at the thoracic spinal cord) and showed enhanced heterogeneity with gadolinium. The lesion was excised through T12-L2 laminotomy. Intraoperatively, the tumor appeared reddish and infiltrative. The frozen section suggested a granulomatous process, while the final pathology confirmed conus medullaris schistosomiasis.

Conclusion: Schistosomal myeloradiculopathy should be considered among the different diagnosis in children presenting with lower thoracic region, conus medullaris, and/or cauda equina infiltrative spinal masses.

Keywords: *Conus medullaris*, Myelopathy, Neuroschistosomiasis, Schistosomiasis, Spinal

INTRODUCTION

Rarely, neuroschistosomiasis presents a parasitic infection of the lower thoracic area, conus medullaris, and/or cauda equina. Notably, schistosomiasis is endemic to South America, Africa, and certain Asian areas and infects at least 200 million people worldwide.[6,10]

Typically, the infection occurs secondary to exposure to water that contains snails with cercariae. The cercariae penetrate the skin and travel through the venous system in which they become adult worms that are capable of releasing eggs. Once in the bloodstream, they are frequently deposited in the gastrointestinal or urinary tract and also rarely in the central nervous system.[1] Due to the presence of valveless venous anastomosis between the pelvic and spinal cord venous plexus, an increase in abdominal pressure may lead to egg migration to spinal cord vessels.[9]

As the parasite reaches the spinal cord, symptoms may appear secondary to the resultant immune response and/or inflammatory process.[6,7,9,10] The clinical presentation and imaging characteristics may mimic tumors, making the establishment of the diagnosis of neuroschistosomiasis challenging even in endemic areas.[3,5]

CASE REPORT

A 4-year-old autistic male from São Paulo presented with a 1-month history of a progressive paraparesis associated with fecal incontinence and urinary retention.

On examination, he was alert, the cranial nerves were intact, and he exhibited a Grade 3/5 motor paraparesis with urinary retention. The thoracolumbar magnetic resonance image (MRI) showed a conus medullaris mass; T2-weighted images demonstrated a T9-T12 syrinx, while the enhanced T1 study showed a heterogeneously enhancing mass [Figure 1].

Surgery and outcome

When the T12-L2 laminectomy was performed, the lesion appeared reddish and infiltrative, simulating a high-grade glioma. The intraoperative frozen sections revealed a chronic granulomatous infectious process. The patient was discharged 5 days later with the same preoperative paraparesis; when the final pathology confirmed conus medullaris schistosomiasis, the patient underwent subsequent treatment with praziquantel (20 mg/kg) [Figure 2]. Three months later, the postoperative MRI showed no residual lesion [Figure 3]. At 4 postoperative months, motor strength in the lower extremities improved to the 4/5 level, and he no longer required a urinary catheter.

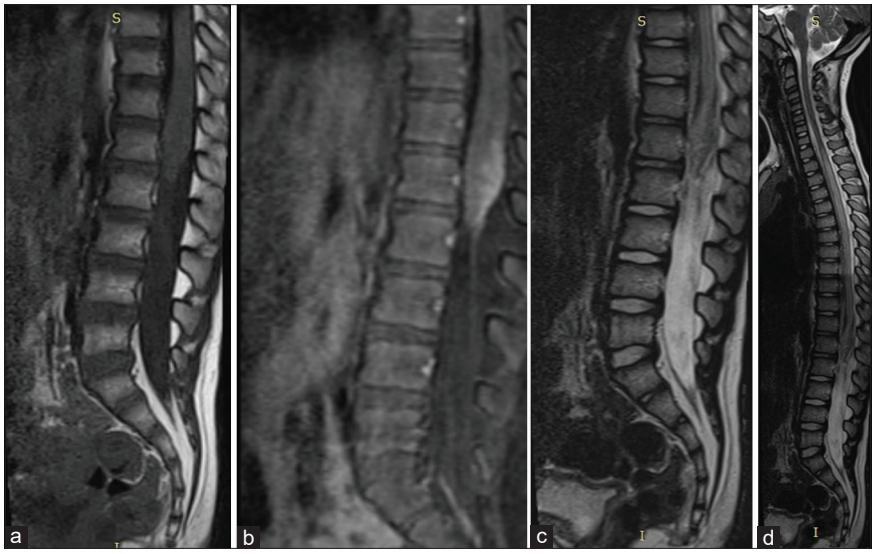

Figure 1: Preoperative magnetic resonance imaging (MRI). (a) Sagittal view of the lumbosacral spine T1-weighted without contrast showing an isointense signal lesion in the conus medullaris. (b) MRI sagittal view of the T2-weighted lumbosacral spine showing hyperintense conus medullaris signal. (c) Sagittal MRI of the lumbosacral T1-weighted spine after gadolinium injection, heterogeneous contrast enhancing in conus medullaris, an apparently infiltrative lesion. (d) Sagittal section of the dorsal column and lumbosacral showing extensive syringomyelia in the thoracic spinal cord.

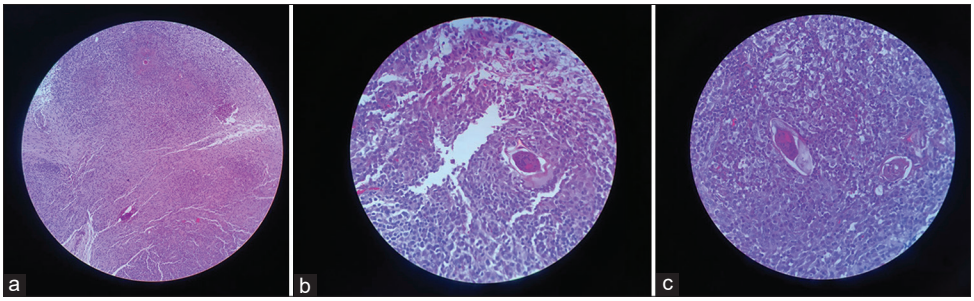

Figure 2: (a-c) Hematoxylin and eosin (H&E) staining showing numerous granulomatous fragments of fibrotic and neural tissue and lymphomononuclear inflammatory infiltrate rich in eosinophils forming granulomatous nodules with necrotic center enclosing parasites with the characteristic spicula of consistent in appearance with *Schistosoma mansoni.*

Table 1: Cases of spinal cord schistosomiasis.

Author	Gender	Age	Level	Removal	Drug treatment
Alsomaili *et al.*	M	24	Thoracic	Biopsy	Praziquantel and steroids
Alsomaili *et al.*	M	29	Lumbar	No	Praziquantel
Salim *et al.*	M	42	Thoracolumbar	Biopsy	Praziquantel and steroids
Salim *et al.*	M	22	Thoracolumbar	Biopsy	Praziquantel and steroids
Salim *et al.*	M	16	Thoracolumbar	Biopsy	Praziquantel and steroids
Salim *et al.*	M	14	Thoracolumbar	Biopsy	Praziquantel and steroids
Salim *et al.*	F	9	Thoracolumbar	Biopsy	Praziquantel and steroids
Mikulich *et al.*	M	28	Thoracic	No	Praziquantel and steroids
Pappamikail *et al.*	M	38	Thoracic	Biopsy	Praziquantel and steroids
Camargo *et al.*	M	48	Thoracic	No	Praziquantel
Kanyamuhunga *et al.*	M	14	Lumbar	Biopsy	Praziquantel and steroids
Labeodan *et al.*	F	10	Thoracolumbar	Surgical removal	Praziquantel
Lighter *et al.*	M	14	Thoracic	No	Praziquantel and steroids
Kim *et al.*	M	25	Conus medularis	No	Praziquantel and steroids
Saleem *et al.*	M	7	Thoracolumbar	Surgical removal	Praziquantel and steroids
Saleem *et al.*	M	9	Thoracolumbar	Surgical removal	Praziquantel and steroids
Saleem *et al.*	M	6	Thoracolumbar	Surgical removal	Praziquantel and steroids
Saleem *et al.*	M	11	Thoracic	Surgical removal	Praziquantel and steroids
Saleem *et al.*	M	27	Thoracolumbar	Surgical removal	Praziquantel and steroids
Saleem *et al.*	M	15	Thoracolumbar	Surgical removal	Praziquantel and steroids
Saleem *et al.*	M	16	Thoracic	Biopsy	Praziquantel and steroids
Saleem *et al.*	M	43	Thoracolumbar	Biopsy	Praziquantel and steroids
Samandouras *et al.*	F	44	Thoracolumbar	Surgical removal	Praziquantel
Paz *et al.*	F	3	Thoracic	No	Oxamniquine and steroids
Paz *et al.*	M	6	Thoracolumbar	No	Oxamniquine and steroids
Paz *et al.*	M	12	Lumbar	Biopsy	Oxamniquine and steroids
Paz *et al.*	M	11	Lumbar	No	Oxamniquine and steroids
Paz *et al.*	M	2	Thoracic	Biopsy	Oxamniquine and steroids
Paz *et al.*	M	14	Lumbar	No	Oxamniquine and steroids
Paz *et al.*	M	10	Lumbar	No	Oxamniquine and steroids

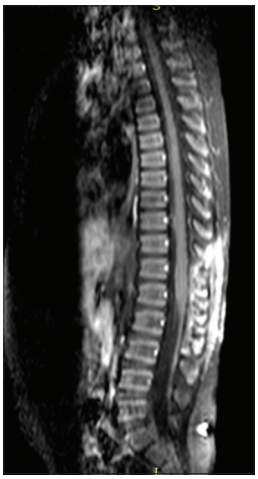

Figure 3: Postoperative magnetic resonance image of the thoracolumbar spine showing the absence of anomalous contrast areas in addition to the absence of recurrence of the lesion.

DISCUSSION

In Brazil, 5.6% of inflammatory myelopathy can be attributed to *Schistosoma mansoni* infection; it must be included among the differential diagnostic considerations for children presenting with acute paraplegia.[1,2,6,9] Typically, schistosomal myelopathy is symptomatic early on, resulting in acute myelopathy and/or radiculopathy.[2,3,6] In this study, our patient experienced the abrupt onset of paraparesis without systemic signs of schistosomiasis. Symptoms are typically attributed to an inflammatory response against schistosome eggs deposited within the conus through the valveless venous plexus of Batson. They result in local tissue edema and/or a granulomatous inflammatory response (e.g., with eosinophils and tissue fibrinoid necrosis around the eggs).[1,4,7,8,10] The differential diagnosis typical includes; an acute vascular event and/or tumor, especially in children from endemic areas for schistosomiasis (Africa and others).[10]

Laboratory studies

Several laboratory tests can assist in establishing the diagnosis of spinal schistosomiasis.[1,9] White blood cell counts may demonstrate nonspecific eosinophilia, cerebrospinal fluid (CSF) findings include lymphocytic pleocytosis, elevated protein, and CSF eosinophilia (64.5%).[1,9] Stool samples

and rectal biopsy also show sensitivities of 50% and 80%, respectively, additionally contributing to this diagnosis.[9,10]

MRI

On MRI, neuroschistosomal infection may mimic intramedullary tumors.[1,2,3,5] The MRI studies in our patient consisted of an expanded/intramedullary spinal cord/conus lesion extending from T12 to L2; it was hyperintense on T2-weighted sequences and heterogeneously enhanced with contrast.

Surgery

Biopsy and/or resection remain the gold standard for establishing the diagnosis of spinal schistosomiasis. However, these can be avoided if there is other sufficient confirmatory evidence.[2,5,7] Notably, the diagnosis is only unequivocally confirmed by direct histopathological evidence (the presence of a Schistosoma ova or worm).[1]

Drug protocol to treat neuroschistosomiasis

The drug protocol for treating neuroschistosomiasis includes praziquantel with corticosteroids; patients on this regimen typically show neurological improvement in 50.0%–60.3% of cases.[2,8] Another alternative medication is oxamniquine [Table 1].[1,4,8]

Outcome

Complete recovery from neuroschistosomiasis is seen in only 30% of *S. mansoni* cases; residual myeloradiculopathy, and other neurological complications attributed to this disease are common.[1]

CONCLUSION

Schistosomal myeloradiculopathy should be considered in children with infiltrative lower thoracic, conus medullaris, and/or cauda equina lesions who present with myelopathy particularly in endemic areas.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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