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Pseudotumoral Form of Cerebral *Schistosomiasis Mansoni*

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Key words
- Cerebral schistosomiasis
- Neuroschistosomiasis
- *Schistosoma mansoni*
- Schistosomiasis

Abbreviations and Acronyms
CT: Computed tomography
MRI: Magnetic resonance imaging

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OBJECTIVE: **To describe published cases of cerebral mansoni schistosomiasis and three others and discuss the diagnosis and treatment of cerebral pseudotumoral schistosomiasis.**

CASE DESCRIPTIONS: **In case 1, a 20-year-old man presented with occipital headache, intense dizziness, visual alterations, nausea, decreased appetite, and asthenia. Cranial computed tomography (CT) revealed an expansive cerebellar lesion in the right hemisphere with no contrast enhancement. The patient had complete resection of the lesion. Anatomicopathological examination revealed a schistosomal granuloma. In case 2, a 22-year-old man presented with generalized tonic-clonic seizure. Cranial CT and magnetic resonance imaging (MRI) revealed an expansive bilateral middle frontal lesion, with contrast uptake close to the cingulate gyrus and corpus callosum. The patient underwent left frontal craniotomy, and an interhemispheric approach was used to resect part of the lesion. In case 3, a 32-year-old man presented with generalized tonic-clonic seizures. Cranial CT showed a hyperdense intense intracranial expansive lesion that presented contrast uptake in the left temporal region. The patient had complete resection of the lesion.**

CONCLUSIONS: **A surgical approach with lesion resection or stereotaxic biopsy is warranted to determine the diagnosis definitively. Antiparasitic drugs must be administered to complete treatment.**

INTRODUCTION

Descriptions of schistosomiasis date back to ancient Egypt. The oldest report of schistosomiasis was found in papyrus texts from 3000 B.C., which was described by Lá Frebvre (28) in 1956. In 1910, Ruffer identified calcified *Schistosoma* eggs in the kidneys of two Egyptian mummies from the 20th dynasty.

Schistosomiasis affecting humans is caused by three species: *Schistosoma mansoni*, *Schistosoma haematobium*, and *Schistosoma japonicum*. The last species tends to produce brain lesions, whereas *S. mansoni* and *S. haematobium* have been associated with spinal cord and root involvement (2, 14, 17, 46, 47, 53, 60, 62, 63).

Schistosomiasis caused by *S. mansoni* is an endemic disease in South America, Africa, the Caribbean Islands, and Eastern Asia. Worldwide, an estimated 200–300 million people are infected with *S. mansoni* (3, 6, 11, 16, 17, 32, 36, 41), and approximately 600 million people are exposed to the disease. In Brazil, 10–20 million people are estimated to be infected with this trematode. *S. mansoni* is the second most com-

mon trematode infecting humans (1, 3, 7, 15, 38, 44, 46, 53).

Because of the small number of cases of the pseudotumoral form of cerebral schistosomiasis mansoni reported in the literature, its physiopathologic mechanism is still poorly understood. The immunologic response against the eggs localized in nervous tissue has been suggested to be the main determining factor of lesions (7, 9, 13, 22, 23, 37, 50, 61, 65).

We discuss the diagnosis and treatment of cerebral schistosomiasis manifesting as a pseudotumor and describe three new cases. In addition, the current literature is reviewed, with reports of all 24 published cases.

CASE DESCRIPTIONS

Case 1

A 20-year-old man, born in the Brazilian southwest, presented with progressive occipital headache associated with significant neck pain. After 1 month, the patient exhibited sudden, intense dizziness, accompanied by alterations in vision, nausea, decreased appetite, and asthenia. Cranial computed tomography (CT) revealed an expansive cerebellar lesion in the right hemisphere with no contrast enhancement. The patient was referred for neurosurgical treatment. Clinical and laboratory tests showed no alterations. Neurologic assessment revealed mild gait ataxia with lateralization to the left and diplopia during right gaze.

The patient underwent right suboccipital craniotomy with complete resection of the lesion in March 2000 (**Figure 1A**). Anatomicopathological examination revealed a schistosomal granuloma containing giant cells filled with *S. mansoni* eggs (**Figure 1B** and **C**).

The patient was treated with praziquantel (20 mg/kg) for 5 days after surgery; no corticosteroids were used as adjuvant treatment. Symptoms improved, and the patient currently reports only a few episodes of intermittent dizziness. The last CT scan, performed in 2004, revealed an area of cerebellar encephalomalacia, with no signs of recurrence. At 8 years of follow-up, the patient remained asymptomatic.

Case 2

A 22-year-old man presented with generalized tonic-clonic seizure. Cranial CT revealed an expansive bilateral middle frontal lesion, with contrast uptake close to the cingulate gyrus and corpus callosum. T1-weighted magnetic resonance imaging (MRI) showed the presence of a bilateral hyperintense lesion with contrast uptake close to the cingulate gyrus (**Figure 2A** and **B**). A stereotaxic biopsy of the lesion had already been performed, but the result was inconclusive.

Because the first biopsy was inconclusive and to improve seizure control, the patient underwent left frontal craniotomy and an interhemispheric approach to resect the lesion in 2000. A hard lesion was observed close to the cingulate gyrus and adhered to the pericallosal arteries. The lesion was partially removed. Anatomicopathological analysis revealed a granuloma containing *S. mansoni* eggs (**Figure 2C**).

The patient was treated with praziquantel (20 mg/kg) for 5 days and long-term phenobarbital. At the last follow-up examination 1 year after surgery, the patient was asymptomatic with epilepsy control; cranial CT scan showed a residual lesion without mass effect.

Case 3

In April 2003, a 32-year-old man presented with generalized tonic-clonic seizures. Cranial CT scan showed a hyperdense intracranial expansive lesion that presented contrast uptake in the left temporal region. Neurologic examination was unremarkable.

The patient underwent left pterional craniotomy with complete resection of the lesion. Anatomicopathological analysis showed the presence of numerous epithelioid granulomas in the cerebral cortex that contained multinucleated giant cells phagocytosing parasite eggs that occasionally exhibited lateral spicules. Intraparenchymatous and meningeal granulomas and a central area of hemorrhage were observed. The final diagnosis was mansoni schistosomiasis in the cortex and meninges. The patient was treated with praziquantel (20 mg/kg) for 5 days and long-term phenytoin.

In the same month, the patient was hospitalized again with febrile symptoms, meningism, vomiting, and leukocytosis (21,700 leukocytes/mm^3). Cranial CT showed mild edema at the site of excision of the lesion. Analysis of cerebrospinal fluid obtained by lumbar puncture showed 2 mg/dL glucose and 1134 cells/mm^3 with 81% neutrophils. Investigation for fungi and cultures were negative. Purulent secretion through the surgical wound was observed. The patient was treated with vancomycin and dexamethasone.

The patient progressed well and was discharged while still taking an anticonvulsant. Brain MRI performed after surgery revealed no residual lesion. At the last follow-up in 2007, the patient was asymptomatic with epilepsy control.

DISCUSSION

Brazil is one of the most important foci of mansoni schistosomiasis in the world, with the disease constituting a serious public health problem (19, 20, 24, 51). It is transmitted by active penetration of cercariae found in infected water into the skin (3, 6, 11, 16, 17, 32, 36, 41). The disease has an acute phase (which passes unnoticed in most cases) and a chronic phase characterized by portal or pulmonary hypertension and nervous system involvement (2, 23, 27, 31). The hepatosplenic form is the most common type (2, 3, 5, 7, 11, 26, 32, 33, 35, 36, 43, 56, 61). Spinal cord schistosomiasis is the most frequent form of nervous system involvement, whereas cerebral schistosomiasis is rare around the world (26, 27, 37, 38, 40, 41, 53, 62, 63).

An anatomicopathological study of autopsy specimens by Pittella (44-46) showed that 26% of patients with the hepatosplenic form of mansoni schistosomiasis presented with brain involvement (11, 25, 44-46). Brain involvement is uncommon because it is the result of an aberrant migration of the worm and eggs, not constituting the pathophysiologic process of the disease. Despite this incidence on autopsy, the clinical form of cerebral schistosomiasis is rare and is limited to certain cases of meningoencephalitis or meningitis and intracerebral hemorrhage and very rare cases of a pseudotumor secondary to schistosomal granuloma formation (1, 8, 15, 16, 43, 44, 46, 48, 50, 53). Most cases with encephalic involvement seem to be asymptomatic. It is unclear why some patients become symptomatic and others remain asymptomatic. An exaggerated inflammatory response would increase the granuloma size and cause a mass effect, leading to symptoms, especially in eloquent areas (45).

As mentioned before, the central nervous system is an unusual site of ectopic infection in schistosomiasis, and the pseudotumoral form is even rarer. Cerebral lesions are caused