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Case Report: Hemianopia: From Suspected Glioblastoma to the Diagnosis of Ectopic Schistosomiasis Haematobium Infection in a Traveler Returning from the Republic of the Congo

Séverine Loridant,^{1†} Gabriel Mouahid,^{2†} Marjorie Cornu,¹ Jean-François Allienne,² Jordan Leroy,¹ Claude-Alain Maurage,³ Emilie Fréalle,¹ Richard Assaker,⁴ Fahed Zairi,⁴ Emmanuel Dutoit,¹ Hélène Moné,^{2*†} and Boualem Sendid^{1*†}

¹Laboratoire de Parasitologie Mycologie, Institut de Microbiologie, Lille University hospital, Lille Inflammation Research International center- Unité Mixte de Recherche 995, Institut National de la Santé et de la Recherche Médicale/Université Lille, Lille, France; ²Centre National de la Recherche Scientifique, Laboratoire Interactions Hôtes-Pathogènes-Environnement, University Perpignan Via Domitia, Institut Français de Recherche pour l'Exploitation de la Mer, University Montpellier, Perpignan, France; ³Laboratoire d'Anatomopathologie, Centre de Biologie Pathologie Génétique, Lille University hospital, Lille, France; ⁴Service de Neurochirurgie, Lille University hospital, Hôpital Roger Salengro, Lille, France

Abstract. Schistosomiasis due to *Schistosoma haematobium* is a widespread disease usually affecting the urinary tract associated with hematuria and kidney disorders. Neurological damage is rarely reported and symptoms are non-specific and may suggest brain tumors such as glioma. We describe the first double ectopic haematobium schistosomiasis case involving the brain and intestine.

A 63-year-old French man was admitted to the emergency department with sudden onset right lateral homonymous hemianopia and mild headache after two similar but transient episodes in the previous 10 days.

On admission, standard vital signs and blood tests were normal except for moderate hypereosinophilia (733/mm³). Ophthalmological evaluation rapidly confirmed the right lateral homonymous hemianopia. Magnetic resonance imaging (MRI) of the brain was performed revealing a left intra-axial occipital lesion suggestive of neoplasia including glioblastoma (Figure 1A and B). An extemporaneous pathological examination of brain biopsies did not reveal a tumor etiology but showed non-caseating necrotizing granulomatous inflammation and the presence of structures with a cuticle evocative of cerebral bilharziasis (Figure 1E). Serological assays revealed a positive hemagglutination test (titer: +1/80) (Fumouze Diagnostics, Levallois-Perret, France), confirmed by enzyme-linked immunosorbent assay (optical density: +2.242) (Scimedx, Denville, NJ), and positive Western blot (three specific bands: 8, 22–23, and 30–33 kDa) (LDBIO Diagnostics, Lyon, France). Microscopic examination of a stool sample by the Ritchie and Junod concentration method and three urine samples after sedimentation and centrifugation were all positive for eggs characteristic of *Schistosoma* spp. Because of the terminal position of the spine, variability in egg morphology, and diversity of localization on morphometric examination, hatching tests and molecular analyses were performed and revealed that few eggs were present in the urine (mean length 136.6 ± 2.4 µm, N = 14), whereas a large number were found in feces (mean length 143.7 ± 1.6 µm, N = 58), and that miracidia hatched from the fecal eggs only. DNA extraction, polymerase chain reaction analyses, sequencing of partial cytochrome C oxidase (CO1, N = 35) mtDNA, and partial internal transcribed spacer-2 (ITS2, N = 39) rDNA, and

phylogenetic analyses were performed as described previously.¹ The molecular analyses revealed only one haplotype of both genes and showed that the parasite recovered from the patient was *Schistosoma haematobium*. The sequences of the CO1 and ITS2 genes have the National Center for Biotechnology Information accession numbers KY967520 and KY967521, respectively.

Additional questioning of the patient revealed a short stay in the Republic of the Congo (Pointe Noire) where he had bathed for 1 hour in the Loukoula River with his wife and friends 4 months ago. All other members of the group were asymptomatic with positive serological tests, but *Schistosoma* eggs were only detected in urine samples collected from three of them. A single dose (60 mg/kg) of oral praziquantel was administered to the case patient and the rest of the group.

One month later, microscopic analyses of urine and stool samples were negative. Magnetic resonance imaging of the brain showed a significant reduction of the lesions at 3 months (Figure 1C and D) and stability at 6 months despite a recurrence of headaches and an increase in visual field defect with visual hallucinations.

This is the first case report to describe double ectopic schistosomiasis haematobium infection: brain and intestine. Neurological damage associated with *S. haematobium* is poorly reported and usually affects the spinal cord with acute transverse myelitis or subacute myelodysplasia of the lumbosacral region.² Brain involvement may occur in the acute phase (acute schistosomal encephalopathy) or chronic phase (cerebral schistosomiasis or pseudotumoral encephalic schistosomiasis). Neuroschistosomiasis can be fatal, especially in its tumor-like form when it affects the cerebellum, but accurate mortality rates are unavailable.³ The availability of neuroimaging, involving computerized tomography scans and MRI is becoming more widespread and may contribute to the early diagnosis of this disease; however, it is nonspecific and may suggest brain tumors such as glioma. From the diagnostic point of view, cerebral lesions associated with schistosomiasis are often difficult to diagnose as definite evidence is mainly established after the onset of nonspecific neurological signs. Moreover, parasitological examination of stools or urine lacks sensitivity and serological assays are not specific for central nervous system (CNS) infection. From the

* Address correspondence to Hélène Moné, University Perpignan Via Domitia, IHPE UMR5244, CNRS, IFREMER, UM, 58 Avenue Paul Alduy, Bât. R, F-66860 Perpignan, France, E-mail: mone@univ-perp.fr or Boualem Sendid, Laboratoire de Parasitologie Mycologie, Institut de Microbiologie, CHRU de Lille, LIRIC-UMR 995 INSERM/Université Lille, 1 Boulevard du Pr Jules Leclercq, F-59045, Lille, France, E-mail: bsendid@univ-lille2.fr.

† These authors contributed equally to this work.

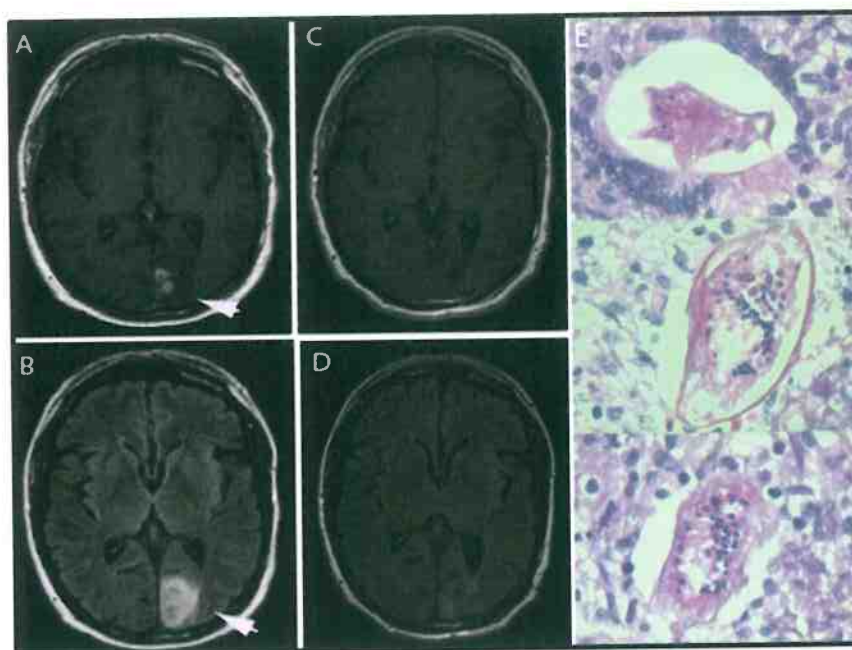


FIGURE 1. Magnetic resonance imaging and histological examination of the brain. (A) Initial appearance of an enhanced lesion surrounded by edema on T1-weighted sequences. (B) Initial fluid attenuated inversion recovery sequences in the left occipital region suggestive of a neoplastic origin. Follow-up imaging revealed disappearance of enhancement (C) and edema (D) at 6 months. (E) Parasitic structures with a cuticle evocative of cerebral bilharziasis on biopsies stained with hematoxylin and eosin ($\times 400$). This figure appears in color at www.ajtmh.org.

therapeutic point of view, symptomatic involvement of the CNS is a serious disease and its outcome largely depends on early treatment with praziquantel.

Diagnosis is difficult in adults and pediatric travelers because of the low parasitic loads during the acute phase of the disease that cannot be observed by microscopy.^{3,4} The low worm burden often makes it impossible to determine the infecting schistosome species in feces or urine.⁴ These difficulties should be circumvented by molecular analyses of either the eggs containing the miracidium or hatched miracidia, if available.⁵

A diagnosis of schistosomiasis should be considered in travelers coming from endemic countries, even if the presentation is atypical. This would avoid unnecessary invasive and potentially dangerous surgical procedures for unrecognized parasitic diseases.⁶ It is difficult to include unusual pathogens in the diagnostic presentations that are incorporated in predefined diagnostic pathways. Gobbi et al.⁷ reported that among seven African migrants, five underwent lung biopsies and were then diagnosed with chronic schistosomiasis and pulmonary nodules due to deposition of schistosome eggs. These authors recommended serological and parasitological screening for schistosome infection in patients coming from areas endemic for schistosomiasis, thereby avoiding lung biopsies and other invasive procedures if the index of suspicion of other severe diseases is low.

In the context of our patient, this screening could have identified schistosomiasis using noninvasive diagnostic methods before neurosurgery. However, the lack of specificity of imaging and the low frequency of these atypical ectopic localizations associated with schistosomiasis haematobium would have required a brain biopsy to prove the involvement of *Schistosoma* in the neurological lesion, on the one hand, and to exclude a neoplasia concomitant

with a parasitic infection, on the other. Diagnosis of atypical clinical forms of schistosomiasis in France is extremely important because this disease continues to spread to new geographic areas, particularly to Europe, associated with the emergence of atypical strains that cause acute schistosomiasis. For example, the recent outbreak of *S. haematobium* found in Corsica^{8,9} showed that the schistosome had a West African origin¹⁰ and was atypical because it had hybridized with a schistosome from cows (*Schistosoma bovis*)^{1,10} and caused acute anuria.¹¹ This case highlights the importance of molecular characterization of strains causing atypical forms of schistosomiasis.

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Authors' addresses: Séverine Loridant, Marjorie Cornu, Jordan Leroy, and Boualem Sendid, Laboratory of Parasitology-Mycology, Lille University Hospital, Lille, France, and LIRIC-UMR 995 INSERM, Université Lille, Lille, France, E-mails: severine.loridant@chru-lille.fr, marjorie.cornu@chru-lille.fr, jordan.leroy@chru-lille.fr, and bsendid@univ-lille2.fr. Gabriel Mouahid, Jean-François Allienne, and Héléne Moné, University Perpignan Via Domitia, IHPE UMR5244, CNRS, IFREMER, UM, Perpignan, France, E-mails: mouahid@univ-perp.fr, allienne@univ-perp.fr, and mone@univ-perp.fr. Claude-Alain Muraige, Laboratoire d'Anatomopathologie, Lille University Hospital Pole Biologie Pathologie Génétique, Lille, France, E-mail: claud-alain.muraige@chru-lille.fr. Emilie Fréalle and Emmanuel Dutoit, Laboratory of Parasitology-Mycology, Lille University Hospital, Lille, France, E-mails: emilie.frealle@chru-lille.fr and emmanuel.dutoit@chru-lille.fr. Richard Assaker and Fahed Zairi, Service de Neurochirurgie, Lille University Hospital, Hôpital Roger Salengro, Lille, France, E-mails: richardassaker@gmail.com and fahed.zairi@chru-lille.fr.

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