



# Enfermedades Infecciosas y Microbiología Clínica

www.elsevier.es/eimc



Diagnosis at first sight

## Hydrocephalus and intracranial hypertension in a woman from Ecuador



## Hidrocefalia e hipertensión intracraneal en una mujer de Ecuador

Javier Nieto-Moragas<sup>a,\*</sup>, Cristina Carrato<sup>b</sup>, Roser García-Armengol<sup>c</sup>, Gema Fernández-Rivas<sup>d</sup>

<sup>a</sup> Department of Clinical Laboratory, Hospital Universitari Germans Trias i Pujol, Universitat Autònoma de Barcelona, Badalona, Spain

<sup>b</sup> Department of Pathology, Hospital Universitari Germans Trias i Pujol, Universitat Autònoma de Barcelona, Badalona, Spain

<sup>c</sup> Department of Neurosurgery, Hospital Universitari Germans Trias i Pujol, Universitat Autònoma de Barcelona, Badalona, Spain

<sup>d</sup> Department of Clinical Microbiology, Hospital Universitari Germans Trias i Pujol, Universitat Autònoma de Barcelona, Badalona, Spain

### Case description

A 40-year-old woman from Ecuador presented to the emergency department (ED) with headache, vomiting, drowsiness, confusion and incoherent speech. She lives in Spain since 2001 and she carries a ventriculoperitoneal shunting since 1993, placed in Ecuador because an obstructive hydrocephalus of unknown origin. In 2013, she consulted repeatedly the ED because hydrocephalus valve mismatches requiring a valve replacement. Laboratory test in peripheral blood showed C-reactive protein level of 34.90 mg/dl and 7800 neutrophils/dl. Cerebro-spinal fluid (CSF) analysis showed 260 red blood cells/ $\mu$ l, 300 leukocytes/ $\mu$ l with a differential of 42% of eosinophils, a protein level of 1.5 g/l and positive xanthochromia. Gram stain of CSF showed no microorganisms and was inoculated onto chocolate blood agar and blood agar plates and incubated in an aerobic atmosphere at 37 °C with negative result after 48 h. Computed tomography (CT) confirms valve dysfunction excluding left ventricular and showed some parenchymal and periventricular calcifications. The patient underwent emergency valve replacement with proximal catheter placement. MRI postintervention (Fig. 1) showed some punctate supratentorial calcifications and two spherical multi-septate lesions with a CSF similar density placed periventricular in the left ventricle. One of the multi-septate lesions supposedly responsible of the symptoms was removed surgically (Fig. 2) and some histological cuts were made of it (Fig. 3).

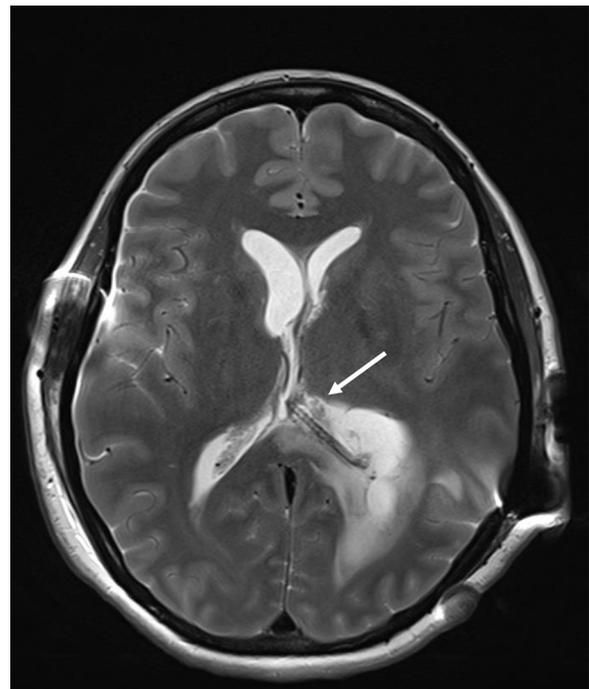


Fig. 1. Magnetic resonance image (MRI) showing a multi-septate nodule (white arrow) producing an expansion of the atrium.

### Evolution

A sample of CSF was sent to the Parasitology Unit of the *Instituto de Salud Carlos III* (Madrid, Spain) to detect by enzyme-linked immunoassay<sup>1</sup> *Taenia solium* antigen and immunoglobulin G (IgG). Both antigen and IgG against *T. solium* tests were positive and it was confirmed with a polymerase chain reaction (PCR) targeted to a *T. solium* DNA on CSF.<sup>2</sup> Parenchymal and periventricular

calcifications, two spherical multi-septate lesions, the presence of anticysticercal antibodies in CSF and been residing in an endemic country, where the patient had already showed intracranial hypertension, lead to the neurocysticercosis diagnosis.

Antiparasitic drugs were not recommended at this stage of the disease and the treatment with parenteral dexamethasone was needed till a progressive neurological improvement. Sequelae

\* Corresponding author.

E-mail address: j.n.moragas@gmail.com (J. Nieto-Moragas).



**Fig. 2.** Macroscopic morphology of a nodular structure extracted surgically.



**Fig. 3.** Photomicrograph of hematoxylin and eosin staining of extracted tissue. A microvillous tegument and a reticular matrix with calcareous bodies are detected.

included language disorder and slow mental activity that improved with neurocognitive rehabilitation.

### Final comment

Differential diagnosis of a cystic brain lesion must contemplate primary neoplasm, metastasis (lung and breast neoplasms are the most common cause), brain abscess, tuberculoma and neurocysticercosis (NCC).

Cysticercosis is defined as the infection caused by the larval stage of *T. solium* once swallowing eggs found in contaminated food, being in contact with human feces of a person who has an intestinal tapeworm or by autoinfection. *T. solium* is a two-host zoonotic cestode and human could be definitive and intermediate host; the last case is when cysticercosis occurs. *T. solium* is found worldwide and parasitosis occurs in regions where humans eat undercooked pork or live in close contact with pigs. NCC is endemic in sub-Saharan Africa, most part of Asia, included China, and Latin America countries. NCC will be developed when oncospheres reach the central nervous system which can be classified in parenchymal and extraparenchymal, depending on the location of the cysts. Seizure is the most common clinical manifestation of parenchymal cysticercosis, caused in 30% of cases by NCC in endemic zones, included Latin America countries.<sup>3,5</sup> Extraparenchymal NCC occurs when cysts are placed in subarachnoid space or in the ventricular system, leading obstruction of CSF flow, producing intracranial

hypertension and hydrocephalus. Mobile extraparenchymal cysts can be the reason of irregular symptoms, such as intracranial hyper/hypotension and sudden death.<sup>3,4</sup> Mononuclear pleocytosis and eosinophilia in CSF could be detected in patients with active inflammation.

In 2017, diagnostic criteria for NCC were edited<sup>5,6</sup> and organized in three categories with many levels of certainty: absolute, neuroimaging and clinical/exposure criteria. Histological confirmation of parasites, evidence of subretinal cysts, and demonstration of the scolex within a cyst are absolute criteria for NCC diagnosis. Cystic lesions without scolex, multilobulated cysts and calcifications are categorized as neuroimaging major criteria. Clinical/exposure criteria include detection of anticysticercal antibodies or cysticercal antigens by enzyme-linked immunoelectrotransfer blot or enzyme-linked immunosorbent assay and residing in endemic areas.<sup>1,6–8</sup> A definitive diagnosis can be made in those having one absolute criteria or two major neuroimaging criteria plus exposure criteria.<sup>5,6</sup>

Patients with extraparenchymal cysts and hydrocephalus secondary to the NCC are candidates to place ventricular shunts or to remove cysts from the ventricles. However, a high mortality exists (up to 50% in two years) related to the number of surgical interventions to change the shunt.<sup>2,3</sup> There is no evidence antiparasitic drugs could contribute to improve calcified cysts resolution. Viable and degenerating cysts can promote a severe immunological response and acute episodes should be treated with corticosteroids (e.g. dexamethasone).<sup>5,6</sup>

### Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

### Conflict of interest

The authors declare that there is no conflict of interest.

### Acknowledgments

Authors thank Silvia Roure and Parasitology Unit of *Instituto de Salud Carlos III* for their work and to guide us through the process.

### References

- Rodríguez S, Dorny P, Tsang VC, Pretell EJ, Brandt J, Lescano AG, et al. Detection of *T. solium* antigens and anti-*T. solium* antibodies in paired serum and cerebrospinal fluid samples from patients with intraparenchymal or extraparenchymal neurocysticercosis. *J Infect Dis.* 2009;199:1345–52, <http://dx.doi.org/10.1086/597757>
- Hernández M, Gonzalez LM, Fleury A, Saenz B, Parkhouse RM, Harrison LJ, et al. Neurocysticercosis: detection of *Taenia solium* DNA in human cerebrospinal fluid using a semi-nested PCR based on HDP2. *Ann Trop Med Parasitol.* 2008;102:317–23, <http://dx.doi.org/10.1179/136485908X278856>
- García HH, Nash TE, Del Brutto OH. Clinical symptoms, diagnosis, and treatment of neurocysticercosis. *Lancet Neurol.* 2014;67:1120–7, [http://dx.doi.org/10.1016/S1474-4422\(14\)70094-8](http://dx.doi.org/10.1016/S1474-4422(14)70094-8)
- DPDx – Laboratory Identification of Parasitic Diseases of Public Health Concern [last reviewed 21.03.17]. Centers for Disease Control and Prevention (CDC) [last visited 21.02.18]; 2017. Available at <https://www.cdc.gov/dpdx/>
- Del Brutto OH, Nash TE, White AC Jr, Rajshekhkar V, Wilkins PP, Singh G, et al. Revised diagnostic criteria for neurocysticercosis. *J Neurol Sci.* 2017;372:202–10, <http://dx.doi.org/10.1016/j.jns.2016.11.045>
- White AC Jr, Coyle CM, Rajshekhkar V, Singh G, Hauser WA, Mohanty A, et al. Diagnosis and treatment of neurocysticercosis: 2017 Clinical Practice Guidelines by the Infectious Diseases Society of America (IDSA) and the American Society of Tropical Medicine and Hygiene (ASTMH). *Clin Infect Dis.* 2018, <http://dx.doi.org/10.1093/cid/cix1084> [in press].
- Wilson M, Bryan RT, Fried JA, Ware DA, Schantz PM, Pilcher JB, et al. Clinical evaluation of the cysticercosis enzyme-linked immunoelectrotransfer blot in patients with neurocysticercosis. *J Infect Dis.* 1991;164:1007.
- Michelet L, Fleury A, Sciuotto E, Kendjo E, Frago G, Paris L, et al. Human neurocysticercosis: comparison of different diagnostic tests using cerebrospinal fluid. *J Clin Microbiol.* 2011;49:195–200, <http://dx.doi.org/10.1128/JCM.01554-10>