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REPORTE DE CASO

Brain and spinal arachnoiditis in a patient with neurocysticercosis: Case report

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Abstract

Introduction: Arachnoiditis is a chronic inflammatory process of the arachnoid layer and subarachnoid space, and many possible etiologies can cause it, including infectious diseases.

Case report: A patient with brain and spinal arachnoiditis was diagnosed and managed on the basis of clinical suspicion as tuberculous meningitis, but when he finished corticosteroids he presented neurological worsening, and after serum western blot immunoassay of *T. solium* positive, a possible racemose neurocysticercosis was diagnosed and treated.

Conclusion: Doctors should consider the importance of nontuberculous etiologies in countries where both neurocysticercosis and tuberculosis are endemic, especially when exist arachnoiditis with cranial basal involvement.

Key words: arachnoiditis, cysticercosis, neurocysticercosis, subarachnoid neurocysticercosis.

Aracnoiditis cerebral y espinal en un paciente con neurocisticercosis: reporte de caso

Resumen

Introducción: La aracnoiditis es un proceso inflamatorio crónico que compromete la aracnoides y el espacio subaracnoideo, y puede ser causada por muchas entidades, que incluyen procesos infecciosos.

Reporte de Caso: Se describe un paciente con aracnoiditis craneoespinal, diagnosticado y manejado, bajo la sospecha clínica como meningitis tuberculosa, pero quien al terminar la terapia con esteroides presentó un empeoramiento neurológico, y posterior a un resultado de western blot para *T. solium* positivo, se diagnosticó y trató como una posible neurocisticercosis racemosa.

Conclusión: Los clínicos deben estar atentos y considerar etiologías no tuberculosas en países donde la neurocisticercosis y la tuberculosis son endémicas, especialmente cuando existe compromiso de las cisternas de la base.

Palabras clave: Aracnoiditis. Cisticercosis. Extraparenquimatosa. Neurocisticercosis subaracnoidea.

Introduccion

Arachnoiditis is a chronic inflammatory process of the arachnoid mater and subarachnoid space, and it could affect the brain, the brainstem, and the spinal leptomeninges[1]. It could be many possible etiologies including infectious diseases, rheumatologic diseases, subarachnoid hemorrhage, non-surgery spine interventions, spinal surgeries, trauma, and cancer related[1].

Neurocysticercosis (NCC), the infection by the larval form (cysticercus) of *Taenia solium*, is the most common helminthic infection of the central nervous system[2]. Humans the sole definitive host carry the adult tapeworm in their intestines (taeniasis), and they could be infected and develop cysticercosis after ingestion of eggs or gravid proglottids expelled with the feces[3-4]. In deficient sanitary conditions, the human feces could be ingested by pigs, the most common intermediate host, and the infective embryos released from eggshells, develop into cystic, fluid-filled larvae o cysticercus with scolex and established in tissues with high vascularity[2].

In the central nervous system, the helminth could lodge the brain parenchyma, subarachnoid space (SAS), intraventricular space, or spinal cord[2-3]. The brain usually precedes the spinal

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involvement, which accounts for 0.7%-3% of all cases, and usually affects the dorsal spine (44.5%) followed by cervical (34%), lumbar (15.5%), and sacral (6%) regions[5]. The spinal infection can arise from: 1) migration of basal subarachnoid cysts through the Magendie or Luschka foramen, 2) hematogenous spread from a gastrointestinal tract, and 3) migration through a dilated ependymal canal secondary to intraventricular hypertension[6]. The incubation period is prolonged, with a median time of 22.2[10–25] years, and 50% of patients could be asymptomatic or present intermittent symptoms and signs[2-3].

We report a patient with brain and spinal arachnoiditis, who was diagnosed and managed as tuberculous meningitis. After neurological worsening, he was readmitted, and a serum western blot immunoassay of *T. solium* was performed, and possible racemose NCC was diagnosed.

Case

A 43-year-old man, a resident of the city of Medellin, Colombia, was remitted from ophthalmology after finding bilateral disc optic swelling. He complaints of five months of episodic, throbbing headache, vertigo, loss of memory, and gait disturbances.

The examination revealed bradypsychia, papilledema, and Romberg. Brain magnetic resonance imaging (MRI) revealed an obstructive supratentorial hydrocephalus with basal meningeal enhancement due to the presence of cyst in the basal cisterns (Figure 1), and cerebrospinal fluid (CSF) analysis showed pleocytosis, hyperproteinorrhachia, and hypoglycorrhachia with an opening pressure of 42cmCSF. Differential diagnosis were negative, and on the basis of clinical suspicion antituberculous and corticosteroids was initiated with dexamethasone being the drug of election starting 20mg for one week, then 14 mg for another week, and then switched to prednisone 50mg which equals 7 mg of dexamethasone.

After finishing the corticosteroids, he was readmitted with headache, gait instability, and lethargy. A second MRI revealed communicating hydrocephalus with cerebral and spinal arachnoiditis (Figure 2), and a Western blot test for T. solium was positive. The patient was managed with the third ventriculostomy, albendazole and corticosteroids but this time starting with prednisolone 75 mg for a week and reducing the dose by 10 mg each week, until reaching 25 mg, which he took for almost five months. Thanks to the good evolution, the costicosteroids dose was decreased by 5mg weekly until he finished the treatment. In addition, hypopituitarism developed, it may be due to sellar involvement, followed by endocrinology.

Discussion

Neurocysticercosis is no longer an exceptional disease in our environment. Health professionals must include it even with initial levels of differential diagnosis algorithms, not only among subjects from endemic areas but also among people who cohabit with them.

The differential diagnosis of this entity in endemic regions, like our patient´s home, is extremely difficult, due to the coexisting endemic tuberculosis and other pathologies, making this case a challenge for the physicians that took care of the patient.

Annular lesions (single or multiple) in the CNS are not specific for neurocysticercosis and represent a diagnostic dilemma, that is why differential diagnosis should include brain abscesses, tuberculomas, neurosyphilis, toxoplasmosis and primary or secondary neoplasms (these may present with similar lesions on CT or MRI and cause mass increase effect with symptoms similar to those of neurocysticercosis).

The clinical manifestations of NCC are not specific and depend on the location of the parasite, the number of cysts, their size, the evolutive stage, and the severity of the immune response of the host[3,7,8].

Parenchymal NCC affects the parenchyma of the brain and the spinal cord, the sulcus of the convexity, and includes the subarachnoid space of the convexity[3]. Seizures (64%) are the most common symptoms, followed by headache[2,4,6]. Generally, it has a good prognosis and presents a complete resolution or calcification after treatment[3].

Extraparenchymal NCC includes: 1) the subarachnoid space (cranial or spinal) the basal cisterns (95/238, 51.6%), Sylvian fissure (23/238, 12.5%), and SAS of medulla (4/238, 2.2%); 2) the ventricular system (62/238; 33.7%); 3) the sellar/suprasellar region; 4) the subdural space and; 5) the optic nerves or retina[3,4].

Subarachnoid NCC is the most serious and difficult form to treat, and 75% of patients do not have a complete response to therapy[3,4]. In this location, the cyst has an abnormal growth of its membrane, losing the scolex, evokes an inflammatory response and causes symptoms related to intracranial hypertension (154/238, 72%), silent lacunar infarcts (28/102, 27.4%), arteritis, mass effect, and arachnoiditis[2,4,6].

The diagnosis of NCC is difficult because clinical features and biochemical laboratory tests could be mimicked by other infections, and histological diagnosis is rarely obtained[2,8].

MRI is the imaging method of choice, and cysticerci has variable appearance according to their degenerative stage, which includes: 1) cystic lesion without enhancement and with mural nodule depicting the scolex "Hole-with-dot" sign (vesicular), 2) cystic or nodular lesions with perilesional enhancement and edema (colloidal and granular-nodular or mixed), and 3) nodular calcified lesion without perilesional edema or enhancement (calcified cysticerci)[2]. Parasites in the SAS or in the ventricular system are often in the cluster group (racemose NCC) or may present as leptomeningeal enhancement (arachnoiditis)[2].

The serological test of choice for identification of anticysticercal antibodies in serum is the electro immunotransfer blot (western blot) assay (EITB)[2,8]. It has a sensitivity of 98% and specificity near 100% in the presence of two or more viable parasites (in the setting of single cyst or calcified parasites 50% may be falsely negative)[2].

Treatment modalities include a medical management (antiparasitic, antiepileptic [AED], and anti-inflammatory drugs) and surgical procedures (cysts resection endoscopically or by open procedure, ventriculoperitoneal shunt placements, and decompressive craniotomies)[2,9].

The best cysticidal agent, the duration of treatment, and the benefit of corticosteroid, methotrexate and etanercept use has not been studied, and the therapy had been based on the medical needs and clinical state[2,9,10] (Table).

The prognosis of extraparenchymal NCC has been improving with the neuroimaging, surgical management, and antiparasitic drugs[9,10]. MRI every three or 6 months is a method used to determine the effectiveness of treatment[10]. The clinical improvement, the CSF parameters, and cestode antigen level guide the intensity and duration of treatment[9,10].

Conclusion

Neurocysticercosis is no longer an exceptional disease in our environment. Health professionals must include it in increasingly initial levels of differential diagnosis algorithms, not only among subjects from endemic areas but also among people who cohabit with them.

The differential diagnosis of this entity in endemic regions is extremely difficult, due to the coexisting endemic tuberculosis and other pathologies such as this case that rule out tuberculosis infection.

Annular lesions (single or multiple) in the CNS are not specific for neurocysticercosis and represent a diagnostic problem. A differential diagnosis should be made with brain abscesses, tuberculomas, neurosyphilis, toxoplasmosis and primary or

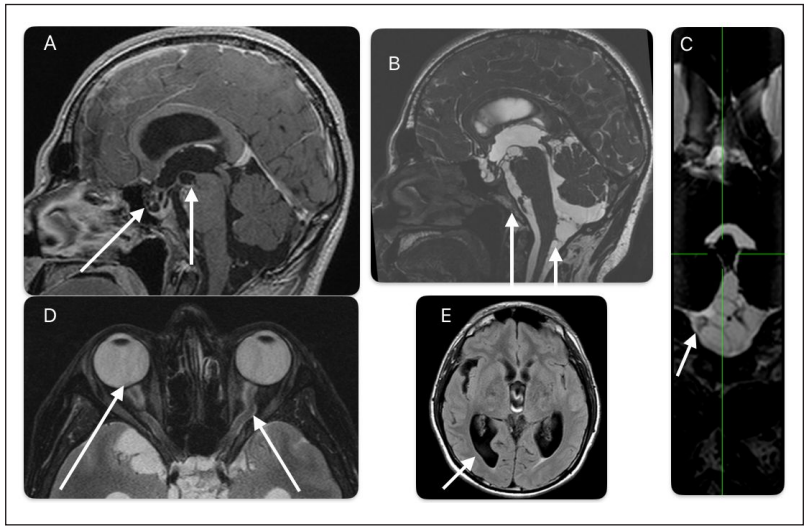

Figure 1. A: Post-contrast sagittal T1 septa with enhancement, cysts and compartmentalization of the subarachnoid space are identified in the interpeduncular, prepontine and suprasellar cisterns.B and C: Sagittal CISS, a decrease in the caliber of the mesencephalic aqueduct is observed in the lower third due to synechiae that generates chronic obstructive hydrocephalus without transependymal migration. In the coronal CISS, cysts are identified in the cisterna magna, prepontine and cervical canal.D: Axial T2 TSE shows tortuosity of the optic nerve with signs of right papilledema due to intracranial hypertensionE: Flair Axial no trasependymal edema identified.

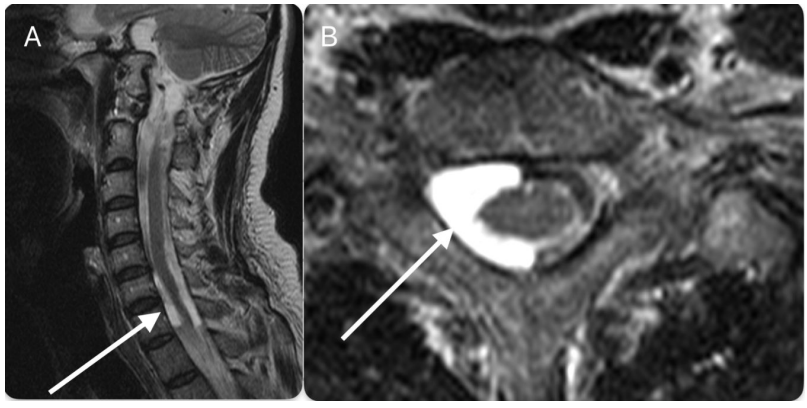

Figure 2. TSE-Axial Cervical-T2 SagittalCompartmentalization of the subarachnoid space in the cervical cord is identified, identifying a cystic, extramedullary, right lateral intradural lesion that surrounds the cord without mass effect at the level of T7.

secondary neoplasms (these can present with similar lesions on CT or MRI and cause mass increase effect with symptoms similar to those of neurocysticercosis).

Table. Summary of treatment recommendations

Parenchymal NCC
Viable cystic lesions and enhancing lesions: Albendazole 15 - 30 mg/kg/day + praziquantel 50 - 100 mg/Kg/day for 10 - 14 days or Albendazole 22 mg/Kg/day for 10 - 14 days when praziquantel is not available Corticosteroids must be used, the optimal dosing has not been established: prednisolone 25 - 60 mg/day or 4 - 10 mg/day of dexamethasone, at least the first 4 - 10 days of treatment. AED for seizures.
Cysticercosis encephalitis: Cysticidal drugs are contraindicated during the acute phase. Control the intracranial hypertension with corticosteroids.
Heavy or refractory infections: Repeat cycles of treatment of 8 - 30 days duration. Corticosteroids are mandatory before, during, and after therapy. Dexamethasone 6 mg/day for 10 days and 8 mg/day for 28 days followed by a 2- week taper. AED for seizures.
Calcified parenchymal disease: Do not need to be treated.
Intramedullary cysticercosis: Albendazole 15 mg/Kg/day for 10 days and Dexamethasone 0.1 mg/kg/day a day before antiparasitic therapy, and during 1–2 weeks, followed by slow taper or, dexamethasone 10 to 16 mg/day with posterior taper, usually after 7 - 10 days, 1 mg/week, or substitute with prednisolone at an equivalent dose 8 - 30 days.
Extraparenchymal NCC
Subarachnoid disease (leptomeningeal) **Racemose Cysticercosis:** Albendazole 20 - 30 mg/Kg/day for 15 - 30 days (repeated cycles may be needed). Praziquantel 50 mg/kg/day may be added. Corticosteroids are mandatory before, during, and after therapy. AED for seizures.
Hydrocephalus or diffuse cerebral edema: No cysticidal drug therapy. Management of elevated intracranial pressure. It often requires a ventricular shunt. Continuous corticosteroid administration (50 mg prednisone three times a week for up two years) may be needed.
Intraventricular disease: **Ventricular cysts:** Do not need cysticidal drug therapy. Endoscopic resection **Angiitis and chronic arachnoiditis:** Do not need cysticidal drug therapy. Corticosteroids mandatory
Cysticercosis of the spinal SAS: Albendazole 20 to 30 mg/Kg/day for 15 - 30 days (repeated cycles may be needed). Praziquantel 50 mg/kg/day may be added. Corticosteroids are mandatory before, during, and after therapy. Surgical Resection.

Adapted of[2,9,10]

Neurocysticercosis should be considered in the differential diagnosis of patients with brain and spinal arachnoiditis. This case illustrates the extremely variable and non-specific nature of the symptoms and signs of NCC. The characteristic neuroimaging findings could be seen in other conditions. Doctors should be taken in account the importance of considering nontuberculous etiologies in countries where both NCC and tuberculosis are endemic, especially when exist arachnoiditis with cranial basal involvement.

Ethical declaration

The authors declare that this manuscript has been written following the Helsinki declaration and has been approved by the ethical committee of the hospital. The authors do not declare any conflicts of interest.

Authors contributions. LG, PB, FH: contributed to the conceptualization of the research. LG, PB, VG: collection of information, construction of the manuscript, review of the bibliography. LG, AR: information collection, analysis and interpretation of diagnostic images.

All authors contributed to, read, and approved the submitted version of the manuscript. Declaration that the content of the article is original

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