Surgical Treatment for Mesial Temporal Lobe Epilepsy in the Presence of Massive Calcified Neurocysticercosis

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Background: Neurocysticercosis (NCC) is the most common parasitic disease of the human central nervous system and a major health problem for most developing countries. The most common clinical manifestations of NCC are epileptic seizures. Whenever epilepsy and NCC coexist in the same patient, an uncertainty may arise about a causal relationship between them.

Observation: We described a female patient with disseminated calcified NCC lesions and intractable epilepsy. Her medical history included cysticercotic meningoencephalitis and status epilepticus caused by active NCC. Fundoscopy showed the ocular presence of parasite; computed tomography of the brain showed evidence of cystic lesions with the scolex and calcified lesions; enzyme-linked immunosorbent assay of the cerebrospinal fluid was positive for cysticercosis. Epileptic seizures started after an 8-year silent period. Magnetic resonance imaging showed left hippocampal sclerosis. Plain x-ray film showed calcifications in muscles and subcutaneous tissue. Video-electroencephalography and ictal and interictal single-photon emission computed tomography disclosed left mesial temporal lobe epilepsy. The patient underwent left temporal lobectomy and has been seizure free since surgery, for a follow-up of 4 years.

Conclusion: This case report highlights and supports surgical therapy in patients with epileptic seizures and calcified NCC, even when there are several calcifications, provided that clear localization of epilepsy has been determined by means of a presurgical workup.

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Neurocysticercosis (NCC) is the most common type of parasitic disease of the human central nervous system, secondary to its infection by the larvae of the tapeworm Taenia solium.1 Neurocysticercosis constitutes a major health problem for most developing countries.2 The main clinical manifestations of NCC are epileptic seizures, headache, and focal neurological deficits. At present, the most reliable tool for diagnosing NCC is neuroimaging by means of computed tomography (CT) or magnetic resonance. On the basis of neuroimaging findings, NCC may be classified into active, transitional, and inactive forms.3 There are 3 possible scenarios of the relationship between NCC and epilepsy, including (1) a noncausal relationship, or simple coincidence of 2 unrelated diseases in the same individual; (2) a causal relationship, ie, cysticercosis as the real cause of focal epilepsy; and (3) dual disease manifestation (commonly referred to as “dual pathology”) in which a coexisting brain lesion (eg, hippocampal sclerosis) and a cysticercotic lesion contribute to the epileptogenesis.1

Definitive evidence of a noncausal relationship is provided when patients with NCC and mesial temporal lobe epilepsy become seizure free after anteromesial temporal lobectomy, without resection of the cysticercotic lesion.4 Causal relationship occurs mainly in those patients with active or transitional forms of intracerebral cysticerci,3 a condition in which seizure incidence is reported to be as high as 91.8%.4 In the inactive forms of NCC, perhaps the most direct evidence implicating calcified cysticercotic lesions as the cause of seizures and other focal neurological signs is the episodic appearance of perilesional edema that may accompany these clinical manifestations.3 This observation suggests that calcified NCC may contribute to the epileptogenic process in some cases. A dual–disease manifestation scenario may occur when calcified NCC is associated with other brain lesions. Epilepsy surgery in such intractable cases may be controversial in light
of reported cases of improved surgical treatment when complete removal of both lesions is performed.\

We herein describe the unique case of a patient with countless calcified NCC lesions who became seizure free after temporal lobectomy. It demonstrates that even extreme dissemination of NCC lesions does not preclude the successful surgical therapy for epilepsy. We believe that this report contributes to the decision-making process about surgical treatment of such patients.

REPORT OF A CASE

A 21-year-old woman with medically intractable complex partial seizures was referred to our center for presurgical evaluation. Medical history included several episodes of cysticercotic meningoencephalitis starting at 6 years of age due to a massive infestation of cerebral parenchyma with cysticerci associated with an intense inflammatory response. These episodes were characterized by headache, vomiting, fever, neck stiffness, and rightsided focal motor status epilepticus. Computed tomography disclosed countless lesions of active (presence of scolex) and transitional forms of NCC. Hydrocephalus and intracranial hypertension due to an intraventricular cyst developed, and a ventriculoperitoneal shunt was then performed. Results of cerebrospinal fluid analysis revealed mononuclear pleocytosis, eosinophilia, reference-range glucose levels, and elevated protein levels. Results of an enzyme-linked immunosorbent assay of the cerebrospinal fluid for cysticercosis were strongly positive. Treatment included oral and intravenous dexamethasone sodium phosphate and phenytoin sodium. The patient also presented with an ocular cysticercus in the subretinal space that caused right monocular blindness. After a silent period of 8 years since her initial seizures, medically intractable complex partial seizures developed.

During the presurgical evaluation, left temporal lobe epilepsy was diagnosed. The CT and x-ray film findings showed disseminated subcutaneous, muscular, and intracranial calcified cysticercotic lesions (Figure, A–C). The magnetic resonance imaging findings also disclosed left mesial temporal lobe atrophy (Figure, D). Results of electroencephalography showed interictal left temporal lobe epileptiform discharges and interictal left temporal intermittent rhythmic delta activity. Results of video-electroencephalographic monitoring showed 100% of interictal spikes on the left temporal lobe and complex partial seizures with ictal electroencephalographic onset in the left temporal region. Results of ictal single-photon emission computed tomography showed a typical hyperperfusion on the left temporal lobe, whereas those of interictal single-photon emission computed tomography showed a moderate hypoperfusion on the same region (Figure, E and F).
We described a female patient who presented with a disseminated distribution of calcified NCC lesions and who underwent successful epilepsy surgery. She initially had cysticercotic meningoencephalitis and focal motor status epilepticus caused by active and transitional forms of NCC. The definitive diagnosis of cysticercosis was determined on the basis of 2 absolute criteria, ie, the detection of the parasite by funduscopic and evidence of cystic lesions showing the scolex on CT findings.

It became clear from the results of the presurgical workup that she had exclusively left mesial temporal lobe seizures, despite the presence of multiple cerebral lesions. The natural history of her epilepsy was indistinct from typical cases of mesial temporal lobe epilepsy, including the presence of an initial precipitating insult, a silent period without seizures, and the late onset of complex partial seizures confirmed by results of video-electroencephalographic monitoring and a seizure-free outcome after surgery. In this case, 2 possible initial precipitating insults were implicated in the pathogenesis of the hippocampal sclerosis, ie, the status epilepticus and/or the meningoencephalitis. The main dilemma during the decision-making process of this case was obviously whether the NCC was related or unrelated to the epilepsy. This definition affected the decision of eligibility or noneligibility for surgical treatment. If the conclusion was that cysticercotic lesions were a coincidental finding, the patient would be a candidate for surgery. On the other hand, if NCC was thought to be causally related to the epilepsy, surgery would not be feasible, considering that removal of all lesions was not possible.

We believe that this case highlights the possibility of successful surgical treatment when patients present with well-defined localization-related epilepsy, despite the existence of disseminated calcified NCC lesions. It is possible that NCC had played a role early as an initial precipitating insult, leading to the development of hippocampal sclerosis. In recent years, however, the presence of calcified NCC and chronic epilepsy was considered to be unrelated in this patient, and calcified NCC per se did not represent an absolute contraindication for the surgical treatment.

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