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Surgical management of cerebral peduncle neurocysticercosis: A case report of decision making in a resource-restricted environment

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ABSTRACT

BACKGROUND: Diagnosis and treatment of neurocysticercosis have always been a challenge, especially in Vietnam, a resource-restricted environment. Here, we reported successful surgical management of cerebral peduncle neurocysticercosis.

PRESENTATION OF CASE: A 65-year-old man with no medical history complained of headache and progressive right-sided hemiparesis for one month prior to admission. On examination, his muscle strength was 3/5 (Frankel grade) with positive Hoffmann and Babinski's signs on the right side. His brain's magnetic resonance imaging suggested a cystic brain tumor (29 × 18 mm) situated in the left cerebral peduncle and hypothalamus region. Enzyme-linked immunoassays (ELISA) of serum for *Cysticercus cellulosae, Echinococcus*, *Toxocara,* and *Amebiasis* were negative. The operation was performed with the modified frontotemporal-orbitozygomatic approach. We extirpated both the cysts. Histopathological examination revealed the larva of *Taenia solium.* Postoperatively, the hemiplegia was completely recovered, and no complication was reported

DISCUSSION: The most recent revised diagnostic criteria of neurocysticercosis emphasised that neuroimaging studies play the most crucial role while clinical manifestations and exposure criteria were only indirect proofs of neurocysticercosis. Neuroimaging and clinical evidence in this patient, however, were more suggestive of cystic brain tumor, which was our most likely preoperative diagnosis. Consequently, total tumor resection required for both histopathological examination and alleviating his hemiplegia.

CONCLUSIONS: Preoperative diagnosis of neurocysticercosis has always been a challenge. Total resection of the cerebral peduncle neurocysticercosis was adequate when the patient had localized neurological deficits.

1. Introduction

Neurocysticercosis (NCC) was the most common parasitic infection of the central nervous system. *Cysticercus cellulosae,* the larval stage of the porcine tapeworm *Taenia solium,* was the pathogen of neurocysticercosis. Cysticercosis was endemic in 60 out of 63 provinces in Vietnam [1,2]. The estimated prevalence of *T. solium* cysticercosis in humans was approximately 13% in rural 'hotspots' [1,2].

In the world, despite many recent revisions of diagnostic criteria, preoperative definitive diagnosis of NCC has always been very challenging due to unfeasible histopathological examinations, atypical clinical and imaging findings, and poor sensitivity or specificity of immunological tests [3]. Until now, neuroimaging studies play a key role in the diagnosis of NCC [3]. Given that high-tech neuroimaging, specific immunological tests, and surgical instruments have not been extensively available in resource-limited settings, the difficulty in diagnosis and treatment of NCC is exacerbated in the context of Vietnam, a lower-middle-income country. Here, we presented the successful management of a 65-year-old male patient with the cerebellar peduncle neurocysticercosis mimicking a cystic brain tumor.

The work has been reported in line with the SCARE criteria [4].

2. Presentation of case

A 65-year-old man with no medical history complained of headache and progressive right-sided hemiparesis for one month prior to admission. He had no nausea, vomiting, and blurred vision. On examination, he was alert and oriented. His vital signs were normal. He had no intracranial hypertension and meningismus. On the

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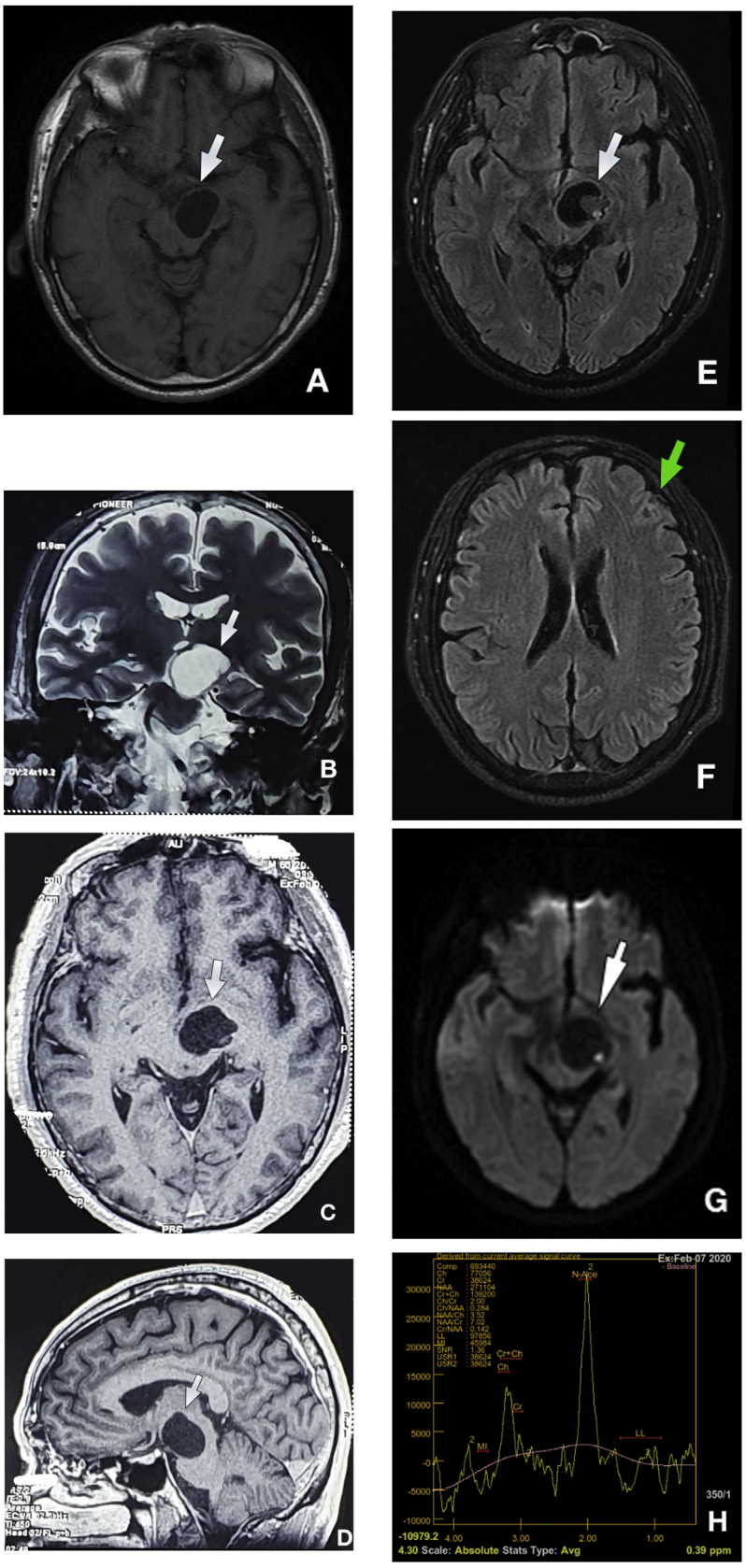

Fig. 1. The magnetic resonance imaging of the brain showed a cerebral peduncle cyst (white arrow) and a suspected lesion at the left frontal lobe (green arrow). (A) T1-weighted, (B) T2-weighted, (C) Axial gadolinium-enhanced T1-weight, (D) Sagittal gadolinium-enhanced T1-weight, (E, F) Axial FLAIR, (G) Diffusion-weighted imaging, and (H) Magnetic resonance spectroscopy.

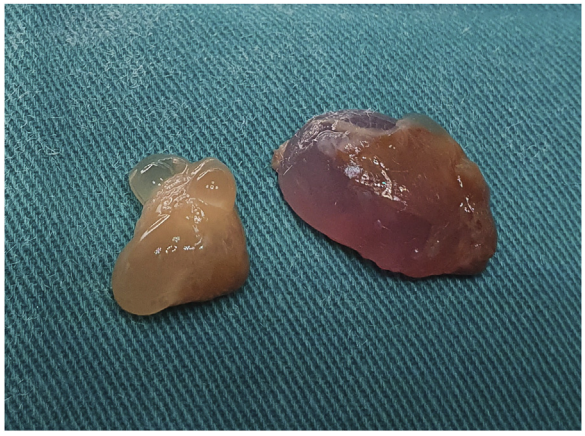

Fig. 2. Gross pathology of neurocysticercosis.

right side, his muscle strength was 3/5 (Frankel grade) with positive Hoffmann and Babinski's signs. Patellar and biceps reflexes were increased. On the left side, his motor and sensory examination were normal. The fundoscopic examination was normal. He denied cranial nerve palsies.

Magnetic resonance imaging (MRI) of his brain showed a 29 × 18 mm, regular, oval thin-walled cyst located at the left cerebral peduncle and hypothalamus region. It was hypointense on T1W and hyperintense on T2W. Inside of the cyst, a superior posterior mass was not contrast-enhancing on gadolinium-enhanced T1W and had restricted diffusion on diffusion-weighted imaging (DWI). On perfusion sequences, the cyst was not hypervascular. On magnetic resonance spectroscopy (MRS), there was no change in levels of Choline and N-Acetyl Aspartate (Fig. 1).

On complete blood count, white cell count was 7.45×10^9 cells/liter, and absolute neutrophils were 6.52×10^9 cells/liter (87.6%) and eosinophils were 0.01×10^9 cells/liter (0.1%). Enzyme-linked immunoassays (ELISA) of serum for *Cysticercus cellulosae, Echinococcus*, *Toxocara,* and *Amebiasis* were negative.

Our preoperative diagnosis was cystic brain tumors (pilocytic astrocytoma, hemangioblastoma, or dysembryoplastic neuroepithelial tumor). Differential diagnoses were neuroglial cyst and neurocysticercosis. The operation was performed to remove the cyst and examine its pathology. The modified frontotemporal-orbitozygomatic (FTOZ) craniotomy and transsylvian approach were chosen. After opening the dura, we exposed a regular, oval thin-walled cyst measuring 1 × 2 cm with clear fluid in the left frontal lobe. We continued dissecting Sylvian fissure to approach the cerebral peduncle. The cerebral peduncle cyst measuring 2 × 3 cm was also well-circumscribed, thin-walled, and intraparenchymal. There was a solid nodule inside the cyst. Both the cysts were extirpated (Fig. 2). Histopathological examination revealed the larva of *Taenia solium.*

Postoperatively, the patient was given Cefoperazone 4 g per day for five days. The hemiplegia was gradually relieved. The patient was discharged after a week. At a three-month postoperative follow-up, the MRI showed no remnant mass, and his muscle strength was fully recovered (Fig. 3)

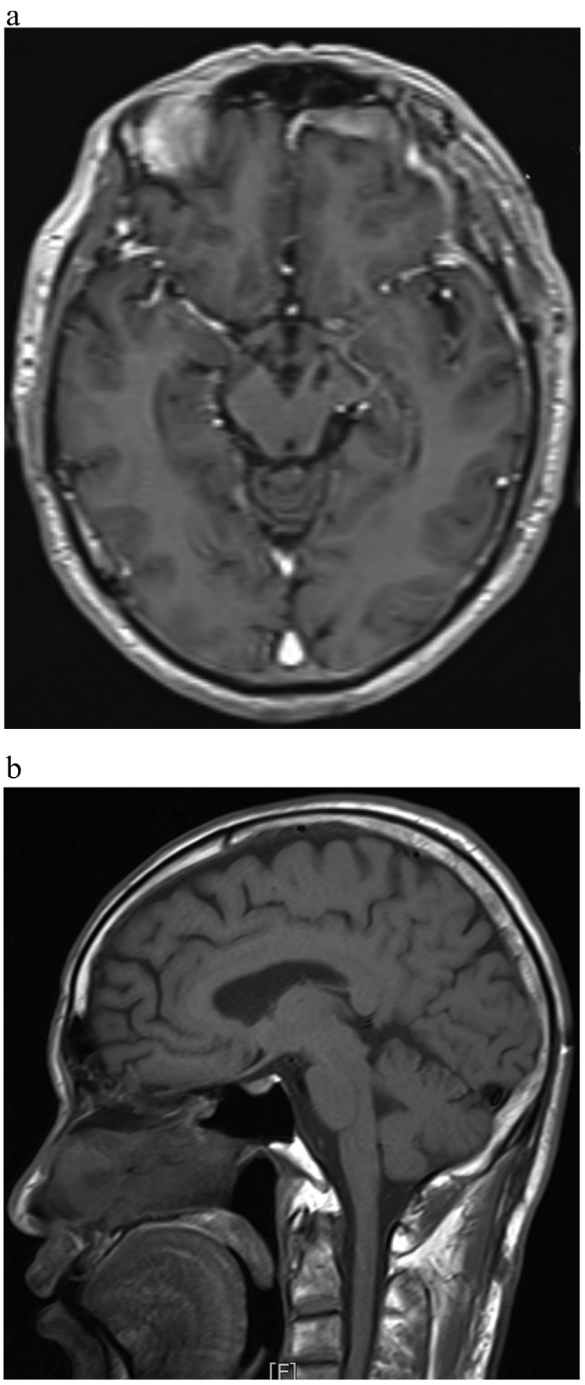

Fig. 3. At a three-month postoperative follow-up, MRI showed no remnant mass.

3. Discussion

The preoperative diagnosis of neurocysticercosis has always been complicated. The most recent revised diagnostic criteria of neurocysticercosis emphasized that neuroimaging studies play the most crucial role while clinical manifestations and exposure criteria were only indirect proofs of neurocysticercosis [3,5]. According to Del Brutto's revised diagnostic criteria (2017), our patient met one major neuroimaging criteria (Cystic lesions without a discernible scolex) and one minor clinical/exposure criteria (individuals coming from or living in an area where cysticercosis is endemic) [5]. Nonetheless, a definitive diagnosis of NCC was still a possibility and should be taken into consideration for lack of conclusive negative tests of anticysticercus antibodies. All negative antibodies to *Cysticercus cellulosae,* in this case, were tested by ELISA but not EIBT, which is the well-standardized immunodiagnostic method for antibody testing in 2017 revised criteria but not extensively available in Vietnam. It should be noted that even if anticysticercal antibodies were positive on EIBT in this case, the definitive diagnosis of NCC could only be made when other etiologies with similar neuroimaging findings were excluded. Neuroimaging and clinical evidence in

this patient, however, were more suggestive of cystic brain tumor, which was our most likely preoperative diagnosis. Consequently, histological confirmation was required for a definitive diagnosis.

Regarding intraoperative decision making, after resection of the frontal cyst with a high likelihood of neurocysticercosis, a question occurred to us when we approached the cerebral peduncle cyst: should it be completely removed or treated conservatively by cysticidal drug therapy. Regarding conservative treatment, one of many cysticidal drugs, albendazole, was safe and effective in reducing the number of cysts and long-term seizure frequency [6], as demonstrated in recent systematic reviews and meta-analyses. However, the potentially harmful effects of cysticidal drug therapy in patients with a large lesion also was stressed [6]. Moreover, the recovery of localized neurological deficits was not reported [7,8]. Therefore, in this case, we ultimately decided to extirpate the cerebral peduncle cyst in order to alleviate his hemiplegia.

4. Conclusion

Preoperative diagnosis of neurocysticercosis has always been a challenge. Total resection of the cerebral peduncle neurocysticercosis was effective when the patient had localized neurological deficits.

Declaration of competing interest

Nothing to declare.

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Ethical approval

The study was approved by the Research Ethics Committee of Hanoi Medical University. The procedures used in this study adhere to the tenets of the Declarations of Helsinki.

Consent

Written informed consent was obtained from the patient and his wife, for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

CRediT author statement

- **He Van Dong:** Conceptualization, Methodology, Investigation, Writing - Review & Editing, Supervision
- **Tam Duc Le**: Conceptualization, Methodology, Investigation, Writing - Original Draft, Writing - Review & Editing, Visualization
- **Dat Tran:** Conceptualization, Writing - Original Draft, Writing - Review & Editing
- **Van Dinh Tran:** Visualization, Resources, Writing - Review & Editing

Registration of research studies

Not applicable.

Guarantor

Tam Duc Le.

Provenance and peer review

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