Case report

Neurocysticercosis presented as a solitary cystic parenchymal lesion mimicking primary brain tumor: A case report

Andreas Soejitno a, I Wayan Niryana b, Ni Putu Sriwidyanic, Ni Made Susilawathi a,*, Ni Putu Witari a, A.A. Raka Sudewia

a Department of Neurology, Faculty of Medicine, Udayana University/Sanglah Hospital, Bali, Indonesia
b Department of Neurosurgery, Faculty of Medicine, Udayana University/Sanglah Hospital, Bali, Indonesia
c Department of Pathological Anatomy, Faculty of Medicine, Udayana University/Sanglah Hospital, Bali, Indonesia

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ABSTRACT

Introduction: Neurocysticercosis (NCC) is an infection of the central nervous system by the larval stage of pork tapeworm (Taenia solium/T. solium). Diagnosing NCC can be challenging, particularly among those who reside in areas with rare occurrence of NCC and atypical manifestation such as a solitary parenchymal lesion. We treated a patient whose initially was diagnosed with brain abscess and later, brain tumor, only finally revealed to be an NCC case.

Case report: A 25-year old male suffered from multiple focal-to-bilateral tonic clonic seizures, was initially diagnosed as brain abscess. He was given antibiotics and anti-seizure medication but the seizure relapsed with a typical semiology. Physical examination demonstrated grade I papilledema, grade 4+ hemiparesis, and headache of vascular origin. Patient was suspected to have oligodendroglioma after underwent head MRI examination and subsequent tumor resection was performed. Pathological anatomy evaluation demonstrated multiple cystic segments containing larva of tapeworm, supporting a diagnosis of active NCC infection. After 14-day course of anthelmintic treatment and resumed AED, patient was seizure-free and NCC was not found upon follow-up CT scan.

Conclusion: NCC, with respect to clinical and radiological manifestations, can be protean. A high index of suspicion towards NCC should always be maintained, particularly among patients originated from endemic area. Appropriate treatment with anthelmintic may result in full disease resolution, thus precluding unnecessary invasive approach.

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Introduction

Neurocysticercosis (NCC) is a zoonotic infection by larva of T. solium in the central nervous system (CNS) [1]. NCC is endemic in South East Asia [2,3], including certain regions of Indonesia (i.e. Bali and Papua), where the majority of the population were non-moslem, therefore has no restriction on pork processing and consumption [4]. The majority (70–90%) of NCC patients presented with seizure which was often focal with or without generalization, and appeared as either solitary or multiple lesions with single or several stages of lesion age upon neuroimaging [5]. Diagnosing NCC can often be challenging when there was only solitary lesion along with the absence of other supporting. The differential diagnosis varies, comprising other intracranial infections such as tuberculosis and toxoplasmosis to malignancy, either primary or secondary brain tumor [6,7]. Herein we reported a patient initially thought to suffer from primary brain tumor who was later discovered to have an active degenerating cyst of NCC with viable T. solium larva.

Case report

A 25-year old male patient from East Nusa Tenggara was admitted to the ER at June 24 2018 with seizure. Patient was complained of having a 2-to-3-minute partial seizure marked by shaking movement of the left extremities preceded by stiffness, then became generalized. During the convulsion, both of patient's eyes deviated toward the left side. This was not the first episode of seizure the patient had. Previously, patient already had seizure twice, with the first episode appeared approximately six months prior to admission. Patient was initially admitted to a different hospital wherein he was diagnosed with brain abscess and given antibiotics and anti-seizure medication. After 3 weeks of treatment, patient was then discharged with continuation of
anti-seizure medication. The seizure relapsed 3 months later. All three seizures had a similar semiology as explained previously.

Upon physical examination, patient was alert and conscious with stable vital signs. Ophthalmoscope evaluation revealed mild papilledema (grade 1). Patient had no cranial nerve palsy but exhibited mild left hemiparesis (grade 4+), particularly on the leg, and mild vascular-type headache. Patient was also assessed with focal-to-bilateral tonic clonic seizure. He subsequently underwent EEG examination but no focal and/or general abnormalities can be found on his recordings.

Head MRI was done and an isointense lesion sized\(2 \times 1.6 \times 1.5\) cm with surrounding focal brain edema in the right frontoparietal cortex was apparent on axial T1 MRI (Fig. 1A), and subsequent gadolinium resulted in the appearance of a rim-enhanced lesion (Fig. 1B, C). Axial T2 MRI demonstrated hyperintensity on the same area (Fig. 1D and E). Consequently, patient was assessed with space occupying lesion on the right frontal cortex, thought to be a brain tumor (i.e. oligodendroglioma) and subsequently underwent craniotomy for tumor resection.

The resected brain tissue demonstrated fibrosis with scattered polymorphonuclear cells, including neutrophils, lymphocytes, plasma cells, and histiocytes. Furthermore, there were multiple cystic segments containing larva of tapeworm (Fig. 1F and G).

Patient immediately received albendazole 400 mg b.i.d for 14 days, with 10 mg of iv dexamethasone t.i.d. tapered off gradually. In addition, patient also received 100 mg of phenytoin p.o. t.i.d since the first day of admission. His seizure did not relapse ever since and he was discharged on the fifth day post resection. Patient was evaluated 1.5 months later with a follow-up head CT scan which demonstrated the absence of both active and/or degenerative lesion.

**Fig. 1.** (A) Axial T1 Head MRI showed an isointense lesion with perifocal brain edema in the right frontoparietal cortex, (B) forming ring-enhanced lesion upon gadolinium administration displayed on axial and (C) sagittal view. (D) T2 axial and (E) sagittal MRI demonstrated hyperintense lesion with surrounding focal brain edema on the same location. (F) gross examination of sectioned tissue revealed glistening cystic lesion (G) microscopic examination demonstrated T.solium larvae complete with its sucker (arrow), surrounded by suppurative inflammatory reaction (100x magnification). (H) A follow-up non-contrast axial head CT scan one month after surgery showed focal hypodense lesion in the post-resected area, suggesting encephalomalacia (arrow) with post-surgical cranial bone defect (arrowhead).
Discussion

NCC is a common infection of the CNS and is known to be the primary preventable epilepsy in various developing countries. Our patient had seizure as his primary symptom before being diagnosed with NCC. He presented a unique case of disease transmission, since he did not consume any porks. However, he did consume various undercooked meats, including horse and ox meat, a rather unusual practice in Indonesia. Currently there has been no documented Taenia infestation among equine or cattle. However, cases of taeniasis and NCC did have been documented in Nusa Tenggara and among resident of Nusa Tenggara who had been diagnosed in Bali [8,9]. According to the latest proposal for a revised diagnostic criteria of NCC, an endemic area was defined as a place where an active transmission has been documented [6]. When referring to our case, patient's hometown was categorized as endemic area in spite of unclear source of intermediate host, thus merit further investigation.

As of neuroimaging, MRI had been proven to be superior over CT scan in detection and characterization of both parenchymal and ventricular NCC [10]. Patient underwent MRI with gadolinium and demonstrated colloidal vesicular stage according to Zhao et al. [10] classification of NCC neuroimaging (Fig. 1B and C). In this stage, larva degenerated from scolex followed by increased viscosity of cystic fluid to become more turbid and proteinaceous, shrinkage but thickening of the cyst wall, surrounded by focal brain edema as a result of ongoing inflammation. Consequently, the cyst becomes mildly and markedly hyperintense when compared with CSF on T1- and T2-weighted images, respectively. In this case, the scolex was not visible anymore, a finding consistent with this stage. However, this stage was still categorized as an active lesion.

The lesion was initially thought to be a primary brain tumor (i.e. oligodendroglioma), given the age and sex of the patient, solitary lesion, location of the lesion (i.e. frontal lobe), and patient's clinical manifestations of focal neurologic deficits [11]. The lesion also appeared slightly hypointense on T1 and markedly hyperintense on T2-weighted MRI (Fig. 1A–E), consistent with typical findings of oligodendroglioma [12]. In fact, NCC cases mistakenly diagnosed as brain tumor was had already been documented [13]. NCC manifested as single cyst in the brain with subsequent surgical resection had been documented previously [7]. A Japanese patient suffered from epileptic seizure had a CT scan demonstrating a 10-mm lesion in diameter with perifocal edema on the left temporal lobe. The lesion was initially thought as brain tumor before the resected tissue displayed a viable cysticercus.

Our patient had eosinophilia approximately two and one month prior to surgery (0.53 and 0.71 × 10^9/L, respectively), indicating parasitic infection among the differential diagnoses. However, he did not have any additional cysticercosis on skeletal survey. In addition, ELISA antibody test against T. solium was undetectable.

Subsequent surgical resection and pathological examination displayed tissue fibrosis surrounded by inflammatory cells and the presence of cystic lesion containing viable larva of T. solium with its visible sucker (Fig. 1G), confirming the diagnosis of an active but degenerating NCC. According to the currently accepted diagnosis of NCC by Carpio et al. [14], our case fulfilled the diagnostic criteria of a definitive parenchymal NCC.

Albendazole monotherapy accompanied with corticosteroid were administered for 14 days post-resection, a regimen based on the current clinical practice guidelines adopted to our hospital protocol [15]. A subsequent follow-up head CT scan conducted one month post surgery displayed no active lesion thereafter (Fig. 1H). Although post-surgical seizure recurrence was relatively low, antiepileptic medication was continued for two years according to our national guideline for the management of symptomatic epilepsy and re-evaluated thereafter for the potential of discontinuation with gradual tapering off.

Conclusion

NCC mimicking a brain tumor could pose a diagnostic challenge, particularly when all other supporting examinations were negative. A differential diagnosis of NCC should always be considered when encountering patients presented with seizure accompanied by solitary cortical brain lesion, particularly of those who came from an endemic area. An appropriate treatment of antihelminthic and corticosteroid were the mainstay of treatment and able to exert full disease resolution.

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Consent

Written informed consent was obtained from the patient 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.

Author contribution

All authors contributed equally in this study.

Conflicts of interest

None.

References

[1] Garcia HH, Nash TE, Del Brutto OH. Clinical symptoms, diagnosis, and treatment of neurocysticercosis. Lancet Neurol 2014;13:1202–15.

[2] Aung AK, Spelman DW. Taenia solium taeniasis and cysticercosis in Southeast Asia. Am J Trop Med Hyg 2016;94:947–54.

[3] Ito A, Li T, Wandra T, et al. Taeniasis and cysticercosis in Asia: a review with emphasis on molecular approaches and local lifestyles. Acta Trop 2019;198:105075.

[4] Sutrisa P, Kapit IN, Wandra T, et al. Towards a cysticercosis-free tropical resort island: a historical overview of taeniasis/cysticercosis in Bali. Acta Trop 2019;190:273–83.

[5] Singh P. Neurocysticercosis. Ther Adv Neurol Disord 2011;4:67–81.

[6] Del Brutto OH, Nash TE, White Jr. AC, et al. Revised diagnostic criteria for neurocysticercosis. J Neurol Sci 2017;372:202–10.

[7] Ito A, Nakao M, Ito Y, et al. Neurocysticercosis case with a single cyst in the brain showing dramatic drop in specific antibody titers within 1 year after curative surgical resection. Parasitol Int 1999;48:95–9.

[8] Wandra T, Ito A, Swaastika K, Darmawan NS, Sako Y, Okamoto M. Taeniasis and cysticercosis in Indonesia: past and present situations. Parasitology 2013;140:1608–16.

[9] Susilawathi NM, Suryaprapra AA, Soejitno A, et al. Neurocysticercosis cases identified at Sanglah Hospital, Bali, Indonesia from 2014 to 2018. Acta Trop 2019;201:105208.

[10] Zhao J-L, Lerner A, Shu Z, Gao X-J, Zee C-S. Imaging spectrum of neurocysticercosis. Radiol Infec Dis 2015;1:94–102.

[11] Engelhard HH, Stelea A, Mundt A. Oligodendroglioma and anaplastic oligodendroglioma: clinical features, treatment, and prognosis. Surg Neurol 2003;60:443–56.

[12] Lee YY, Van Tassel P. Intracranial oligodendrogliomas: imaging findings in 35 untreated cases. AJR Am J Roentgenol 1989;152:361–9.

[13] Bouillet S, Monteil P, Dauthieres M, Rouget A, Guerin J, Vital A. Two cases of neurocysticercosis mimicking brain tumor. Ann Pathol 2003;23:355–7.

[14] Carpio A, Fleury A, Romo ML, et al. New diagnostic criteria for neurocysticercosis: reliability and validity. Ann Neurol 2016;80:434–42.

[15] White Jr. AC, Coyle CM, Rajeshkhar V, 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;66:e49–75.