CASE REPORT

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The diversity of neurocysticercosis clinical manifestations in Bali, Indonesia: a case series



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Abstract

Background Neurocysticercosis (NCC) is a parasitic disease of the nervous system caused by the larval form of *Taenia solium* which is started to be neglected despite its high prevalence in poor and developing countries. Neurocysticercosis has various clinical manifestations and radiological findings ranging from parenchymal to extra parenchymal location, thus can make it difficult to diagnose.

Case presentations We report four cases of NCC with varying manifestations, including epilepsy, chronic headache, space-occupying lesions with increased intracranial pressure, and asymptomatic case. NCC diagnosis requires imaging studies with either a head CT-scan or head MRI. Management of NCC is based on the clinical manifestations extended from antiepileptic drugs, analgetics, and antiedema to surgery besides anti-parasitic albendazole.

Conclusions This case series describes the clinical manifestations of NCC and the radiologic findings and treatment specifically related to parenchymal and extra-parenchymal lesions.

Keywords Clinical manifestations, Case report, Neurocysticercosis, Parenchymal, Extraparenchymal

Introduction

Neurocysticercosis (NCC) is the most common parasitic infectious disease of the central nervous system (CNS) caused by the larval form of the *Taenia solium* (*T. solium*). Clinical manifestations of this zoonotic and food-borne disease is pleomorphic from asymptomatic, mild to severe symptoms that may extend from different degrees of disability up to death [1-3]. *T. solium* larvae in the CNS can be found in the brain parenchymal or extraparenchymal areas (intraventricular, subarachnoid, and spinal cord) causing various neurological manifestations including epilepsy, chronic headache, hydrocephalus,

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high intracranial pressure (ICP), stroke, and cognitive impairment [1, 4, 5].

Case presentations

We report four cases of NCC with pleomorphic clinical manifestations and provided the results from serology and imaging studies. The summary of our case presentations is depicted in Table 1. Head CT scan in this case report used GE revolution, type XR C203J, 16 slices, manufactured from France. We used Siemen brand, Magnetom Skyra type, 3 Tesla from Germany for the MRI examination. Serological examination was carried out by Enzyme-Linked Immunosorbent Assay (ELISA) from serum samples using the Multiskan Go spectrophotometer, Thermo Fisher Scientific produced by Finland at the Department of Parasitology, Faculty of Medicine, Udayana University, Bali.

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Tal	ble 1 Su	ummai	ry of cá	Table 1 Summary of case series						
No	No Patient M/F	t M/F	Age	Origin	History of pork consumption	Clinical findings	Serology Imaging	Imaging	Management	Outcomes
-	<	Σ	4	Bali Island	Yes	-Uncontrolled general tonic clonic epilepsy in the last 7 years with irregular treat- ment history using phenytoin	Positive	Head CT scan: multiple calci- fications on both hemisphere (Fig. 1)	-Albendazole 400 mg q.12.h per oral for 1 month -Dexamethasone 10 mg q.6.h intravenous for 3 days then tappered down -Phenytoin 100 mg q.8.h per oral	Controlled seizure during treat- ment
7	Ф	ш	28	Timor Island	Yes	-Chronic throbbing headache for 3 months - Normal funduscopy result	Positive	Head MRI: multiple vesicular lesions with perilesional edema, vesicular lesion with scolex (Fig. 2)	-Albendazole 400 mg q.12.h per oral for 1 month -Methylprednisolone 16 mg q.8.h per oral tapering off for 2 weeks	Clinical improvement dur- ing treatment
ω	U	Щ	40	Bali Island	Yes	-Progression of chronic severe headache, nausea, vomit- ing, double vision, wobbly walking, and history of seizure in 3 months Bilateral papilledema and paresis of bilateral abdu- cens nerve -Cerebellar syndrome with impaired coordination	Positive	Head CT scan: obstructive hydrocephalus with suspected infratentorial mass (Fig. 3) Head MRI: intraventricular cyst in 4th ventricle with eccentric nod- ules (scolex) (Fig. 4)	-Dexamethasone 5 mg q.6.h intravenous for 3 days then tappered down -Ventriculo-peritoneal shunt placement -Albendazole 400 mg q.12.h per oral for 1 month after the VP-shunt placement	Clinical improvement follow- ing the VP-shunt placement and medication
4	۵	Σ	41	Bali Island	Yes	Chronic right knee pain with- out neurological complaint	Negative	Head CT scan: multiple calcifications in the right frontal area Genu X-ray AP/lateral: cigar- shaped multiple oval lesions suggesting in tramuscular cysticercosis (Fig. 5)	-Sodium diclofenac 50 mg q.12.h per oral -Albendazole 400 mg q.12.h per oral for 2 weeks	Pain improvement

M male, *F* female

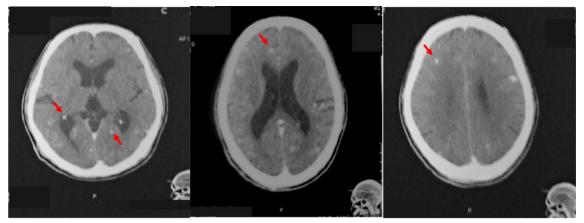


Fig. 1 Imaging of patient A; contrast-head CT scan showed multiple calcifications on both hemispheres

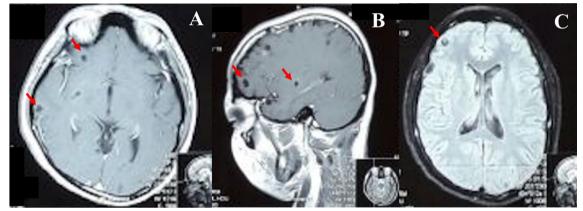


Fig. 2 Imaging of patient B; Head MRI with T1 contrast on axial (A) and sagittal view (B) showed multiple vesicular lesions with perilesional oedema; T2 with FLAIR (C) showed vesicular lesion with scolex

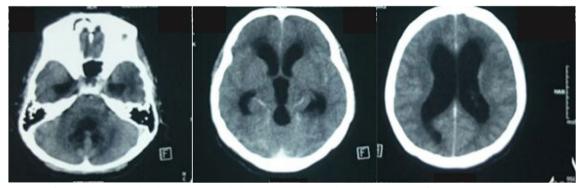


Fig. 3 Imaging of patient C; contrast-head CT scan showed obstructive hydrocephalus with suspected infratentorial mass

Discussion

Four cases above describe the various symptoms and radiological findings on parenchymal (epilepsy, chronic headache, and asymptomatic) and extraparenchymal (cerebellar syndrome with high-pressure obstructive hydrocephalus) NCC. Diagnosing neurocysticercosis requires absolute criteria (based on histopathology, the presence of subretinal cysticercosis or scolex on

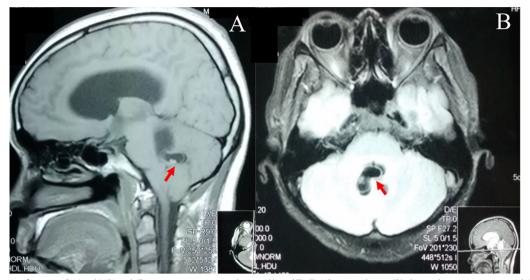
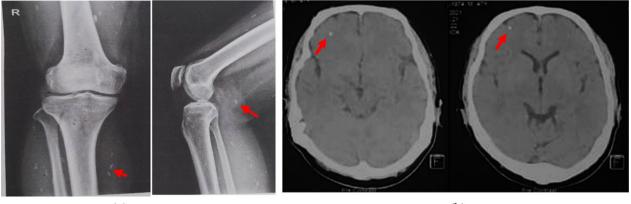


Fig. 4 Imaging of patient C; Head MRI with T1 contrast on a sagittal view (A) and T2 FLAIR on axial view (B) showed cystic mass around the 4th ventricle area with eccentric hyperintense nodule as scolex (red arrow)



(a)

(b)

Fig. 5 Imaging of patient D, a. Right genu AP/lateral X-ray showed multiple oval-shaped lesions around the intramuscular area with a "cigar shape" appearance; b Contrast head CT-scan showed multiple calcifications on cortex and subcortex of right frontal lobe

neuroimaging), neuroimaging criteria (major, confirmatory, and minor), and clinical/exposure criteria (major and minor), leading to definitive or probable NCC [2].

All cases were definitive NCC: Patients A and D met 1 major neuroimaging criteria (calcifications in brain parenchyma) and 2 clinical criteria (epilepsy in endemic areas, positive serology in patient A and intramuscular cysticercosis in patient D originating from regional endemic), while patients B and C met only 1 absolute criteria (scolex on neuroimaging). Ventriculo-peritoneal (VP) shunt was performed on patient C. All patients received albendazole and symptomatic medications with good adherence and showed improvements in their signs and symptoms including headache, seizure, and focal neurological deficits. Patients were from Bali and Timor Island which are endemic areas of NCC, where pig farming and raw/undercooked pork consumption are common [2, 3]. Active *T. solium* transmission in Bali is limited to the slopes of Mount Agung with poor environment and sanitation, [3, 6-8] and a lot of the community seek work in the downtown area, increasing the risk as a taeniasis carrier and transmission of NCC [3, 9].

Neurocysticercosis clinical manifestations vary from asymptomatic to severe critical symptoms [5, 10, 11] associated with larval size, number, stage, location, and host immune response [4, 5, 12]. The highest predilection of NCC is in the brain parenchyma particularly in the grey and white matter junction [5, 11], causing adult-onset epilepsy in endemic areas. Headache is reported in one-third of cases and becomes the second most common presentation after epilepsy [13, 14]. Chronic tension- or migraine-like headache may appear as an isolated symptom or part of the high ICP syndrome [12, 13], or related to the cyst degeneration causing inflammation and pain sensation [14]. Asymptomatic neurocysticercosis with calcification accounts for 5–25% [4], whereas at autopsy, it is estimated to occur in 50% of NCC cases [11–13]. Neuroimaging studies found more than one-third of asymptomatic NCC cases [12].

Extraparenchymal neurocysticercosis is a malignant form of NCC found in the intraventricular, cistern/subarachnoid, spinal, and ocular areas [15, 16] with worse symptoms and prognosis. Common clinical manifestations of extraparenchymal neurocysticercosis are obstructive hydrocephalus by larvae blocking the ventricular system or communicating hydrocephalus due to basal subarachnoid space inflammation [11, 11, 17, 18]. The oncosphere from the larvae penetrates the intraventricular/subarachnoid area through the choroid plexus causing inflammation or space-occupying lesions in the area, especially in the fourth ventricle, which may cause brainstem syndrome [15].

Neuroimaging with head CT-scan is more commonly being conducted than MRI in our setting. The calcifications of parenchymal of NCC can be best visualized using the head CT-scan compared to head MRI. On the other hand, the diagnosis of extra parenchymal NCC, especially in the intraventricular and cistern areas, is very difficult to establish using a head CT scan, since it appears isodense with cerebrospinal fluid (CSF), hence the head MRI is an option in the diagnosis of extra parenchymal NCC with the most common vesicular stage [17].

Management of parenchymal NCC consists of symptomatic therapy utilizing antiepileptic drugs (AED), antiinflammatory, analgesics, and antiparasites [4, 11, 13]. Headache management is tailored according to the type and the underlying cause [13]. Corticosteroids in NCC counter the inflammatory process of natural cyst degeneration or the result of antiparasitic drug administration [13, 18]. Antiparasitic drugs (albendazole, praziquantel) destroy active or degenerating cysts. Combination with steroids is recommended for active cysts. The VP shunt procedure treats high-pressure hydrocephalus due to obstruction in the ventricular area, although in some cases brain cyst removal is required [5, 11, 13].

There are some strengths and limitations to this case report. These case series provide a wide range of clinical manifestations of NCC, from asymptomatic to neuroemergency cases. The endemicity areas of NCC are shown not only from Bali Island but also from Timor Island. Besides, we also displayed complete aspects of Del Brutto's criteria to diagnose NCC from this case series including the absolute, neuroimaging, clinical, and exposure criteria. Unfortunately, no data are available on longterm follow-up of clinical and radiological outcomes, since patients were referred to their primary care.

Conclusion

Neurocysticercosis is known as "the great imitator" due to various neurological manifestations; therefore, clinicians are required to consider neurocysticercosis in diagnosing patients with seizure, headache, and spaceoccupying lesion with high ICP in the endemic area.

Abbreviations

NCC	Neurocysticercosis
CT	Computed tomography
MRI	Magnetic resonance imaging
CNS	Central nervous system
ICP	Intracranial pressure
ELISA	Enzyme-linked immunosorbent assay
Μ	Male
F	Female
VP	Ventriculo-peritoneal
AP	Antero-posterior
FLAIR	Fluid attenuated inversion recovery
CSF	Cerebrospinal fluid
AED	Antiepileptic drug

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Author contributions

NMS acted as the principal investigator and provided conception, design, and technical guidance for all aspects of this project. IAAP, AAAS, VTD contributed equally to the manuscript conception, writing, and editing for submission. IMS, KS, and AARS contributed to supervise and review the data. All authors read and approved the final manuscript.

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Availability of data and materials

The patient data described in this case report are not publicly available due to patient privacy concerns but may be available from the corresponding author (susilawathi@unud.ac.id) upon reasonable request.

Declarations

Ethics approval and consent to participate

Our study was approved by Institutional Review Board of Faculty of Medicine Universitas Udayana/Rumah Sakit Umum Pusat Sanglah Denpasar No.715/ UN14.2.2.VII.14/LT/2021 on March, 9th 2021. Written informed consent to participate from each subject were available.

Consent for publication

The patients described in this report have provided written informed consent for participation and publication.

Competing interests

The authors declare that they have no competing interests.

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