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Case report

Acute disseminated encephalomyelitis complicating dengue infection with neuroimaging mimicking multiple sclerosis: A report of two cases

S. Viswanathan[a,*], N. Botross[b], B.N. Rusli[b], A. Riad[c]

[a] Department of Neurology, Kuala Lumpur Hospital, Kuala Lumpur, Malaysia
[b] Johor Bahru Clinical School, Monash University, Johor Bahru, Malaysia
[c] New Castle University Medicine Malaysia(NUMed), Johor Bahru, Malaysia

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ABSTRACT

Acute disseminated encephalomyelitis (ADEM) complicating dengue infection is still exceedingly rare even in endemic countries such as Malaysia. Here we report two such cases, the first in an elderly female patient and the second in a young man. Both presented with encephalopathy, brainstem involvement and worsening upper and lower limb weakness. Initial magnetic resonance imaging (MRI) of the brain was normal in the first case. Serum for dengue Ig M and NS-1 was positive in both cases. Cerebrospinal fluid (CSF) showed pleocytosis in both with Dengue IgM and NS-1 positive in the second case but not done in the first. MRI brain showed changes of perpendicular subcortical palisading white matter, callosal and brainstem disease mimicking multiple sclerosis (MS) in both patients though in the former case there was a lag between the onset of clinical symptoms and MRI changes which was only clarified on reimaging. The temporal evolution and duration of the clinical symptoms, CSF changes and neuroimaging were more suggestive of Dengue ADEM rather than an encephalitis though initially the first case began as dengue encephalitis. Furthermore in dengue encephalitis neuroimaging is usually normal or rarely edema, haemorrhage, brainstem, thalamic or focal lesions are seen. Therefore, early recognition of ADEM as a sequelae of dengue infection with neuroimaging mimicking MS and repeat imaging helped in identifying these two cases. Treatment with intravenous steroids followed by maintenance oral steroids produced good outcome in both patients.

1. Introduction

Dengue infection is an arboviral infection transmitted by the Aedes aegypti mosquito, caused by 4 distinct viral serotypes(DEN-I to DEN-4). DEN-2 and 3 serotypes are usually associated with neurological complications which include encephalopathy, encephalitis, strokes, immune-mediated syndromes (acute disseminated encephalomyelitis (ADEM), myelitis, Guillain–Barré syndrome), neuromuscular and ophthalmological complications (Carod-Artal et al., 2013; Puccioni-Sohler et al., 2013, 2012; Gupta et al., 2013; Domingues and Kuster, 2014). Dengue induced ADEM is still quite rare even in endemic areas, so early recognition and identification with neuroimaging can improve overall morbidity and mortality. Here we retrospectively report two cases of ADEM presenting as a sequelae of Dengue infection and discuss their unique clinical features and neuroimaging.

2. Methods

We retrospectively identified two cases of ADEM preceded by Dengue infection from 405 cases of idiopathic inflammatory demyelinating diseases at two Malaysian tertiary referral centers. Informed consent was obtained from the patients or their caregivers for publication purposes. Ethical approval was obtained from the medical research ethical committee within the Ministry of health Malaysia for the purpose of publication of this case report.

2.1. Case 1

A 61 year-old Indian lady, well prior to admission presented with a three day history of fever, lethargy, arthralgia, myalgia and diarrhea. On day 4, she developed sudden onset of urinary incontinence with altered mental status, generalized body weakness rapidly becoming comatose requiring intensive care. Examination revealed, normal vital signs with Glasgow coma scale (GCS) of Eye 1, Motor 2 (flexion to pain) and verbal 1 (4/15). Pupils were equal and reactive. Initially, both upper and lower limbs had reduced tone, intact reflexes and down going plantar responses. The rest of the systemic examination was within normal limits.

* Corresponding author.
E-mail address: shivenda70@yahoo.com (S. Viswanathan).

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Hemogram showed normal hemoglobin, erythrocyte sedimentation rate, white cell counts with thrombocytopenia; platelet count of 54,000/mm^3. Urine analysis, chest Xray and blood cultures were unremarkable. Dengue panel tested positive for Non-structural 1 viral antigen (NS1), Dengue IgM and IgG antibodies. Lumbar puncture was clear, colorless, normal in opening pressure with polymorphs of 67%, mononuclear cells of 33%, normal glucose and proteins. Cerebrospinal fluid (CSF) for latex agglutination and cultures were negative for bacteria, fungi, herpes and mycobacteria tuberculosis. Infective screens for mycoplasma, leptospirosis, legionella, herpes and varicella were all negative. EEG done showed excess of slow waves of theta and delta range,with bursts of generalized pseudoperiodic discharges suggestive of an encephalopathy with electrographic status epilepticus. MRI of the brain done within the first week of illness failed to show any abnormalities. She was treated for possible dengue encephalitis with intravenous Ceftriaxone,Dexamethasone, Acyclovir and antiepileptics for 2 weeks. However she failed to improve neurologically and her GCS remained poor at 4/15. Repeat lumbar puncture revealed high CSF protein of 1.0 g/mm^3, acellular with normal glucose and negative CSF oligoclonal bands as were the repeat cultures. CSF NS1 and dengue IgM were not done. Repeat MRI brain (T2 weighted imaging (WI)/ Flair/DWI) showed wide spread asymmetrical flocculent brainstem, fronto-temporal, parieto-occipital subcortical periventricular hyperintense lesions of varying sizes mimicking multiple sclerosis and hypointensities on T1 weighted imaging. (Fig. 1, a-d) Gadolinium T1 imaging was not done. A diagnosis of ADEM complicating dengue encephalitis was made..

She was started on intravenous methylprednisolone one gram /day for five days followed by tapering doses of oral prednisolone 1 mg/kg. Gradually, her conscious level improved. She began to verbalize with slow improvement in motoric power at 6 weeks. With Intensive inpatient physiotherapy and rehabilitation, the patient gradually improved cognitively over 2 months, able to obey commands, and walk short distances with support. Post discharge, she was lost to follow-up.

2.2. Case 2

A 30 year old male, Malay lecturer was admitted with history of fever, myalgia, joint pains, poor oral intake of 2 weeks duration with nausea, headache, vomiting and diarrhea. Initially on admission,the patient's conscious level was full, vital signs showed evidence of tachycardia with temperature of 37.8 °C with normal blood pressure. Blood investigations revealed low total white cells of 3700 cells/mm^3, thrombocytopenia of 60,000, normal hemoglobin and ESR, with Dengue panel for IgM and NS1 positive. Over the subsequent 3 days, his platelet count started to recover. On day 4 of admission, his platelet

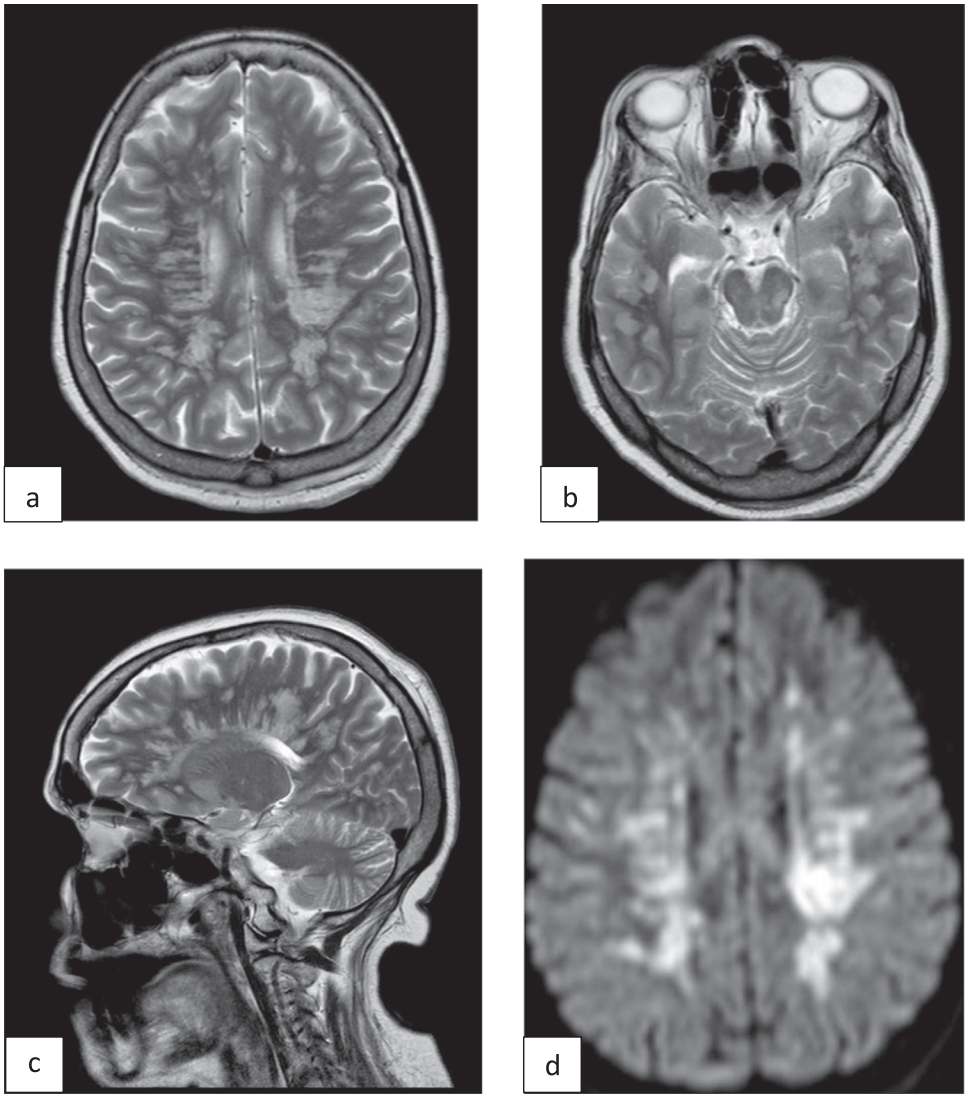

Fig. 1. a) Axial T2WI showing diffuse periventricular white matter lesions, b) Axial T2W1 showing midbrain and temporal lobe white matter lesions, c) Sagittal T2WI showing periventricular dawson finger like white matter lesions in ADEM mimicking MS, d) Axial DWI showing bilateral symmetrical periventricular lesions.