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Research Article

An Uncommon Presentation of a Common Disease - Dengue Presenting as Hemorrhagic Acute Disseminated Encephalomyelitis

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Abstract

Dengue fever is a major seasonal health care problem in the tropics and the developing countries like India affecting thousands of patients every year. The common presentations include fever, myalgia, arthralgia and sometimes shock and bleeding manifestations. Acute disseminated encephalomyelitis (ADEM) complicating dengue infection is rare even in endemic countries such as India. We report a case of dengue fever presenting as ADEM.

Keywords: Dengue; Mosquito; Acute Disseminated Encephalomyelitis (ADEM)

Introduction

Dengue infection is caused by 4 distinct virus serotypes (DEN-I to DEN-4), which are closely related antigenically. It is an arbovirus infection transmitted by the Aedes aegypti mosquito [1-3]. The epidemiology of dengue cases has shown a rise with the passing years number of dengue cases has been rising steadily [4]. In 2018, the total number of cases reported was more than 80,000 with 134 deaths [5]. 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 [6-10]. Although still rare (1 - 5% of dengue cases), neurological involvement has been increasingly reported in dengue epidemics. Furthermore, the figures can be misrepresenting the burden as many cases may be underestimated [11].

ADEM following dengue infections is very infrequent and very few cases have been documented. We present a case of ADEM following dengue infection.

Case

A 20-year-old male presented to the emergency with a fever of 3 days duration and altered sensorium following an episode of generalised tonic-clonic movements of the body, for 1 day. Fever was low grade, continuous, and not associated with chills. On examination, the patient was hyperventilating and unresponsive, Glasgow Coma Scale was 7 (E2V2M3). His pulse was 122 beats per minute and blood pressure was 100/60 mm Hg. Respiratory and cardiovascular system examinations were normal. Abdominal examination showed hepatomegaly 2 finger breadth below the costal margin. On neurological examination, the patient had neck rigidity. Pupillary reflexes were normal. The rest of the systemic examination was unremarkable. The patient was catheterised and supportive treatment including antiepileptics and parenteral fluids were given.

Examinations revealed a hemoglobin: 15.4 g/dl, hematocrit: 46%, white cell count: 11,600/mm3, with polymorphs 53% and lymphocytes 46%, platelet count: 80,000/mm3, blood urea of 26 mg/dl, serum creatinine: 0.6 mg/dl, alanine aminotransferase: 38 U/L, and aspartate aminotransferase: 95 U/L. S. Na+ 132 meq/l, S. K+ 4.5 meq/l and S. Ca2+ 9.1 mg/dl. Cerebrospinal fluid examination showed no cells and normal protein (58 mg/dl) and normal glucose (83 mg/dl; blood glucose was 110 mg/dl).

Radiological examinations revealed a normal chest X-ray and a Non-Contrast computed tomography head was done in the Emer-

gency Department. Subsequently, Magnetic resonance imaging (MRI) brain (Figure 1) was suggestive of diffuse white matter oedema suggestive of ADEM.

In further investigations, the NS1 antigen and IgM antibody were positive for dengue. Ona neurological advice, high dose Pulse steroid therapy was also given for 3 days. Within 48 hours the patient started showing improvement with GCS improving to 15/15. He was discharged after 6 days of admission.

Figure 1: MRI showing diffuse white matter oedema suggestive of ADEM.

Discussion

Acute disseminated encephalomyelitis (ADEM) presents as an acute-onset neurological dysfunction following a medical event such as an infection or vaccination [12]. The annual incidence of ADEM is reported to be 0.4 - 0.8/100,000 and the disease more commonly affects children and young adults, probably related to the high frequency of exanthematous and other infections and vaccination such as measles-mumps-rubella (MMR), polio, diphtheria-pertussis-tetanus (DPT), influenza, human papillomavirus, hepatitis B, rabies, and Japanese B encephalitis, but no definitive conclusions can be made about the association of ADEM, and a specific vaccine in this age group [13-17,27,28]. In dengue infection, the pathophysiology of neurologic manifestations may be related to direct viral invasion; systemic complications related to dengue infection such as stroke and hypokalaemic paralysis, or immune-mediated, autoimmune reaction secondary to dengue infection [18-22]. Patients present with neurological dysfunction resulting from autoimmune reaction causing several areas of white matter brain lesions and imaging with MRI shows white matter lesions in the centrum semiovale, corona radiata, thalamus, extensive involvement of the frontal, parietal, and temporal white matter as well as the corpus callosum. White matter lesions also involved the brainstem, bilateral thalami, and cerebellum. Demyelinating lesions with or without foci of haemorrhage on MRI are probably pathognomonic of ADEM following dengue infection. The pathological hallmark of ADEM is perivenular inflammation with demyelination of neurons usually in the white matter [23]. Clinical evaluation, magnetic resonance imaging, and cerebrospinal fluid study are most useful in establishing the diagnosis and ruling out important differential diagnoses. Corticosteroids are the mainstay of treatment and the role of other modalities of treatment, such as plasma exchange and intravenous immunoglobulin are based on the similarity of pathogenesis of ADEM with that of MS [24,25]. Neuroimaging in dengue associated ADEM may mimic multiple sclerosis-like lesions. Prognosis is generally good in ADEM with excellent functional recovery in 90% of cases [17,26].

Conclusion

In conclusion, neurological involvement in dengue though rare can occur. It is important to identify these neurological features through history, dengue serology in endemic areas and neuroimaging with repeat studies where necessary as early diagnosis and successful treatment with steroids improves the outcome. The dissimilarity between the clinical and radiological findings in Dengue associated ADEM exists and repeat brain neuroimaging may be needed. There is a rising trend in dengue fever and dengue hemorrhagic fevers cases with increased morbidity and mortality which prompted us to report this uncommon yet fatal outcome. A high index of suspicion must be kept for early recognition of these potential complications, especially if a patient presents with associated neurological symptoms.

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