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Reversible Jack-o'-Lantern Sign in Postdengue Hemorrhagic Encephalitis: A Rare Phenomenon

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Dear Editor,

Dengue is caused by an arbovirus belonging to the Flaviviridae family. It has four serotypes: Dengue virus (DENV) nonstructural (NS1) antigen to DENV NS4 antigen. DENV NS2 and NS3 are most frequently implicated in the neurological manifestations that have been reported to occur in 0.5−7.4% of symptomatic cases.[1] Though classically considered a nonneurotropic virus, neurological complications are commonly seen in dengue. We report a case of 28-year-old female with postdengue hemorrhagic encephalitis in whom full clinical recovery occurred after treatment, including reversal of the neuroimaging pattern seen in dengue.

A 28-year-old female presented to us with a 2-week history of fever and generalized body aches, and 1 week later she had developed diplopia, dysphagia, dysarthria, and altered sensorium. There was no rash, seizures, visual complaints, history suggestive of connective-tissue disorders (e.g., lupus, systemic sclerosis, or rheumatoid arthritis), or any other significant medical history. Prior to the present admission, an evaluation performed elsewhere showed thrombocytopenia (89,000/μL), and ELISA revealed positivity for NS1 antigen. She was drowsy but arousable by painful stimuli upon arrival at the hospital. Her pupils were equal in size and reactive, while extraocular movements revealed bilateral restricted abduction, left lower motor neuron facial palsy, brisk reflexes, and bilateral extensor plantars. A provisional diagnosis of acute disseminated encephalitis (ADEM) was considered.

A further evaluation revealed hemoglobin at 13.5 g/dL, total leucocyte count of 3,400/μL, and platelet count of 119,000/μL. A liver function test showed elevated aspartate transaminase at 583 IU/L (normal=0−41 IU/L), alanine transaminase at 1,435 IU/L (normal=0−49 IU/L), bilirubin at 0.68 mg/dL, albumin at 4.17g/dL, sodium at 131 mmol/L, and potassium at 3.71 mmol/L, while renal function tests produced normal results. A cerebrospinal fluid (CSF) analysis produced normal results, with polymerase chain reaction revealing negativity for dengue in the CSF, while the serum was positive for IgM dengue but negative for IgG dengue, repeat NS1 antigen, and Japanese encephalitis virus and chikungunya virus. Ultrasonography findings for the abdomen were normal. However, brain T2-weighted and fluid-attenuated inversion recovery MRI revealed symmetrical hyperintensities in the bilateral thalamus, pons, midbrain, and cerebellum (Fig. 1A-F). Patchy diffusion restriction in the pontine lesion gave the appearance of an atypical jack-o'-lantern sign due to selective involvement of tracts in the pons. In a typical jack-o'-lantern sign there is sparing of corticospinal and corticobulbar tracts forming the eyes, and teeth are formed by the spared medial leminiscus and spinal trigeminal tracts. However, in our patient there was relative sparing of specific tracts in the pons, such as the right corticospinal tract being spared so that only one eye of the jack was visible, the medial leminiscus being involved only on the right side, and additionally both medial longitudinal fasciculi being involved to form the teeth, which were completed laterally by sparing of the lateral leminiscus and spinal trigeminal tract. In

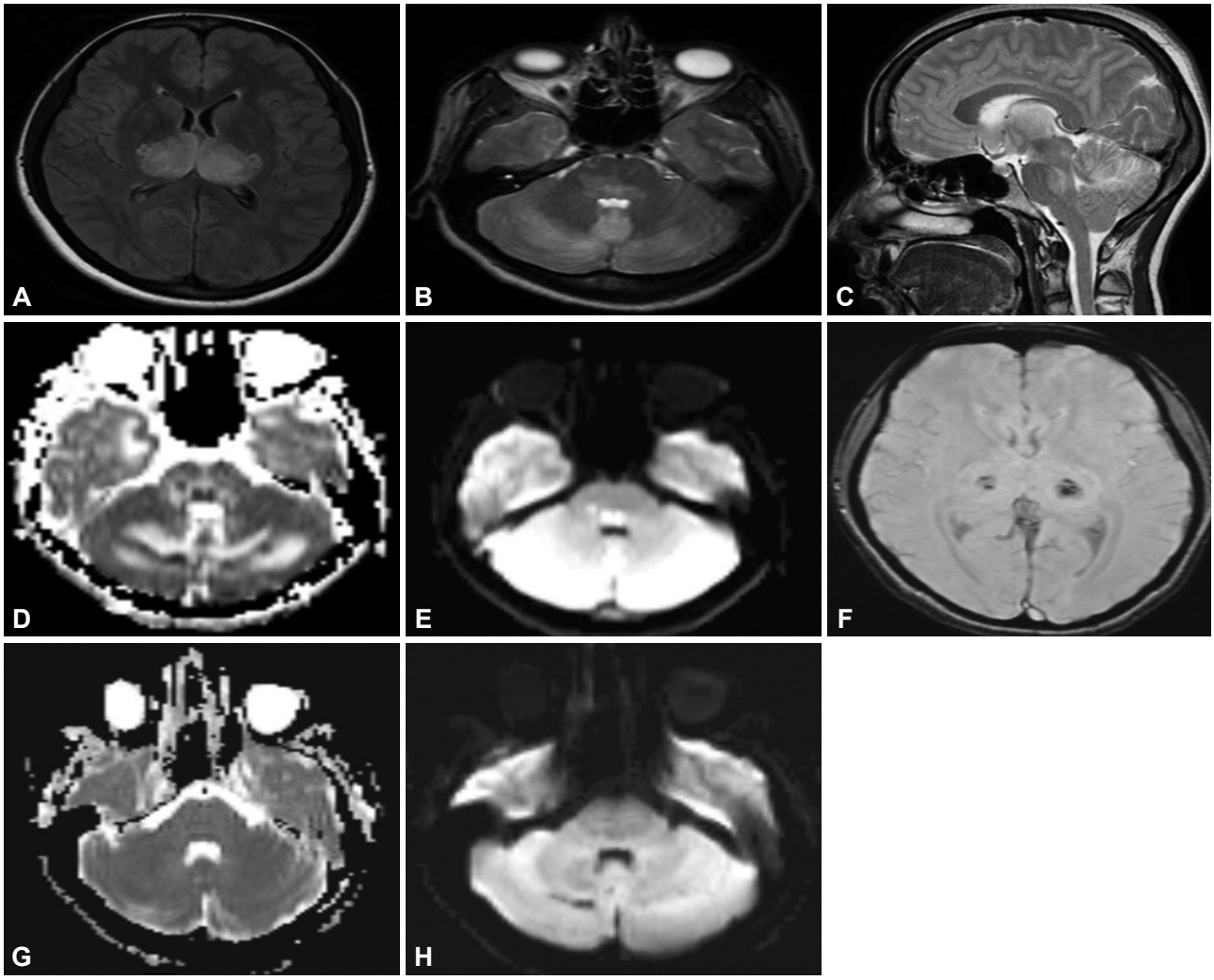

Fig. 1. Brain MRI findings. A and B: Axial fluid-attenuated inversion recovery/T2-weighted images showing symmetric hyperintensities in bilateral thalami and patchy involvement in the pons. C: Sagittal T2-weighted MRI showing hyperintensities in the midbrain, pons, and superior cerebellum. D and E: ADC map and DWI show patchy diffusion restriction in the pons forming an atypical jack-o'-lantern pattern. F: Susceptibility-weighted imaging shows evidence of blooming in bilateral thalami. G and H: In follow-up MRI after 3 months, DWI with the corresponding ADC map showed resolution of the pontine lesion and disappearance of the lantern sign. ADC: apparent diffusion coefficient, DWI: diffusion-weighted image.

addition, there was evidence of blooming on susceptibility-weighted images in both thalami suggestive of a double doughnut sign. There was minimal enhancement of the lesions in the thalamus in contrast imaging.

A final diagnosis of postdengue hemorrhagic encephalitis was made. The patient was managed with intravenous methylprednisolone at 1 g for 5 days followed by maintenance oral steroids, in addition to supportive symptomatic treatment. After 3 months of follow-up there was significant improvement in the symptoms of the patient. She was ambulatory without neurological deficits and repeat imaging showed that the lantern sign had disappeared (Fig. 1G and H).

The spectrum of neurological manifestations was classified by Murthy[2] into three categories. Neurological complications related to the neurotropic effect of dengue virus have been explicitly detailed by Carod-Artal,[1] which include immune mediated syndromes such as ADEM, myelitis, neuritis, and Guillain-Barré syndrome. Our patient presented with acute hemorrhagic encephalitis, which is an uncommon but severe variant of ADEM that has a fulminant course. The pathological hallmark is perivenular inflammation with demyelination and necrotizing vasculitis of venules.[3]

Neuroimaging findings in dengue vary. They may be normal, but hemorrhages, cerebral edema, and focal abnormalities involving the basal ganglia, hippocampus, and thalamus can be seen. In addition, extensive lesions involving the midbrain, cerebellum, thalamus, and medial temporal region have been reported.[4] Brain MRI in our patient showed the involve-

ment of the thalamus, cerebellum, midbrain, and pons in a rare pattern known as the jack-o'-lantern sign; while this has been reported previously, the sign in our patient was not typical.[5] The selective pattern of involvement of particular brain structures gives characteristic imaging patterns, which may help in differentiating it from other viruses, although the specificity and sensitivity of this sign for dengue encephalitis are not known. Appropriate management can be effective, with the overall mortality rate ranging from 3% to 5%. Cases of encephalitis with widespread involvement of the brainstem are usually fatal, as seen in a previous patient who exhibited a lantern pattern.[5] Glucocorticoids are effective in treating postinfectious encephalitis,[3] which is why an early diagnosis is crucial for a good prognosis; this was also the case in our patient, who recovered completely. Full clinical recovery after steroids with reversibility of the lantern sign was a significant finding in our case, which has not been reported previously.

Author Contributions

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Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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