Myoclonus as a late manifestation of West Nile disease

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DESCRIPTION

A 68-year-old man presented with rhythmic shaking of right-sided extremities that occurred several times per day. The patient appeared confused with incoherent speech. His medical history was significant for West Nile fever diagnosed 8 weeks prior and managed supportively. During that admission, there had been extensive testing that was negative for Human T-Lymphotropic Virus (HTLV), HIV, syphilis or antinuclear antibodies. On examination, there was involuntary myoclonic jerking of the left face, neck and arm (online supplementary video 1). MRI of the brain was notable for leukoaraiosis (figure 1).

Electroencephalography (EEG) showed continuous generalised slowing, suggestive of a moderate diffuse encephalopathy. There was no evidence of electrographic seizures or epileptiform activity. Testing of the blood and cerebrospinal fluid (CSF) did not show any signs of meningitis. CSF culture and testing for herpes simplex virus were negative, as was MRI of the entire spinal cord. Supportive treatment of myoclonus included using baclofen 10 mg three times daily and intermittent boluses of lorazepam. A trial of levetiracetam 750 mg twice daily was also attempted but did not result in significant change.

After 5 days, there was a marked reduction in myoclonic jerking, but it was still evident (online supplementary video 2). By day 7, there was resolution of myoclonic activity and the patient was now able to follow simple commands and talk, but he appeared to now have a flaccid paralysis of the left arm. On day 9, the patient began to move the left arm, but there was now increased tone and cogwheel rigidity of the right arm. By day 11, this resolved. Repeat EEG during the episode of flaccid paralysis showed no evidence of electrographic seizures or epileptiform activity. During this time, the patient had much improved mentation and was alert and fully oriented. Given the clinical picture with normal mental status and consciousness, combined with EEG and MRI, we thought it unlikely that these episodes were focal seizures. Thereafter, the patient had a steady recovery with normal mental status and motor examination.

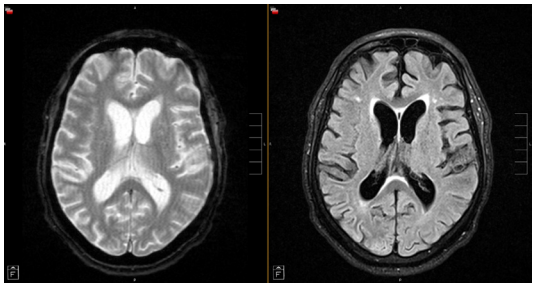

Figure 1 Areas of hyperintense fluid-attenuated inversion recovery (FLAIR)/T2 foci noted in the superficial and deep subcortical white matter of the supratentorium.

Learning points

- West Nile virus is well known to cause meningitis and acute flaccid paralysis, but less is known about the delayed motor symptoms and clinical outcomes of West Nile fever.
- Myoclonus of the upper extremities and face can be a late presentation of West Nile infection.
- The long-term outcomes of dyskinaesias in West Nile infection are not well defined but can be favourable as in this case, with supportive care.

West Nile virus (WNV), a member of the Japanese encephalitis virus antigenic complex, can lead to a variety of clinical symptoms ranging from a mild influenza-like illness to neuroinvasive disease. Advancing age increases the risk of neuroinvasive disease.[1] Patients with WNV encephalitis have been reported to develop movement disorders including tremor, myoclonus and parkinsonism.[1 2] As demonstrated, there was myoclonus of the upper extremity and facial muscles that persisted even during sleep. The sequelae and long-term prognosis of WNV infection is not well understood, but myoclonus of the upper extremities and face has been reported to persist for months.[2] In this case, the motor symptoms resolved with supportive care, indicating some patients with WNV encephalitis can have a favourable course.

Contributors SM and BB provided direct patient care and conceived the idea for the report. KS and SC performed the literature search and drafted the discussion. SM and BB wrote the case report. SM and SC undertook revisions of the manuscript. All authors contributed to and approved the final manuscript.

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