Japanese encephalitis (JE) is caused by the JE virus (JEV), a flavivirus with five known genotypes, which is transmitted between water birds and/or pigs by vector mosquitoes. Humans are dead-end hosts of the virus, and the infection manifests as inflammation of the central nervous system. The virus has been detected across a wide area covering eastern and southern Asia, northern Australia, and Papua New Guinea and is responsible for approximately 68,000 clinical cases annually (1,2).

JE virus infections in humans are mostly asymptomatic and less than 1% of infections result in clinical disease, with the severity ranging broadly from mild febrile illness to acute meningomyeloencephalitis. JE has been associated with a variety of neurological abnormalities, such as altered sensorium, seizures, focal neurological deficit, and acute flaccid paralysis (AFP). However, to date, AFP has never been reported as an initial manifestation of JE. Here, we present a case of AFP manifesting as the initial symptom of JE in a Chinese patient. A 30-year-old Chinese man was admitted to the West China Hospital of Sichuan University after experiencing AFP in the right upper limb, followed by hyperpyrexia and unconsciousness. Assay of cerebrospinal fluid from a lumbar puncture revealed high levels of proteins and anti-JE virus IgM antibodies. Intravenous acyclovir was administered; however, the weakness persisted and more extensive muscle wasting from the proximal to distal right upper limb occurred over 7 months. This case report highlights that JE needs to be added to the differential diagnosis of AFP in adults, especially in JE endemic seasons and areas.
in the lower limbs, positive nuchal rigidity, and Kernig sign. Routine blood test showed a high white blood cell count (12×10^9 /L, 4–10×10^9 /L) and a high neutrophil ratio (90%). The remaining laboratory tests for liver and kidney function, electrolytes, coagulation function, thyroid function, tumor markers, immune function, and so on were normal. Anti-JEV IgM antibody was detected by enzyme-linked immunosorbent assay (commercial kit) in the serum and cerebrospinal fluid from lumbar puncture. Cerebrospinal fluid testing showed a high number of cells (100×10^6 /L) and proteins (0.94 g/L) and the remaining tests, such as chloride, glucose, and smear for fungi, bacteria, and mycobacterium were normal. Electromyography indicated neurogenic damage in the upper limbs, predominantly in the right upper limb, and probable injury of the anterior horn (right C5-T1, left C5-6). No significant alterations were observed in magnetic resonance imaging of the brain and cervical spinal or water imaging of the right brachial plexus. These results led to a diagnosis of JE with myelitis. Intravenous acyclovir, along with rehabilitative and supportive therapies, was administered for 12 days; however, the weakness persisted. At the 7-month follow-up after discharge, the patient showed similar muscle weakness as at discharge but with more extensive muscle wasting from the proximal to distal right upper limbs.

To our knowledge, this is the first case report of a JE patient with AFP as the initial clinical manifestation. Previous studies suggest that a third of JE patients show generalized or focal weakness, without atrophy, involving primarily the upper motor neuron units, and that a quarter of patients show muscle wasting involving hands, feet, and flexors of the neck (8–10). A previous study suggested that the anterior horn cell was involved to varying degrees in many JE patients, which contributes to focal or general weakness and muscle wasting (11). In our patient, JEV infection manifested with AFP in the right limb, which is consistent with previous reports that JE patients can present symptoms of focal weakness and muscle wasting.

Some studies have reported that AFP presents in JE patients, but never as the initial manifestation (12–14). This case highlights that JE, as well as Guillain Barré syndrome, myasthenia, and acute transverse myelitis, need to be added to the disease list for differential diagnosis of AFP in adults.

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Conflict of interest None to declare.

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