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Case Report

Japanese encephalitis virus associated post-infectious relapsing acute onset chronic demyelinating polyradiculoneuritis

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ABSTRACT

Chronic inflammatory demyelinating polyradiculoneuropathy (CIDP) is an uncommon manifestation of Japanese encephalitis (JE) virus infection. JE is a neurotropic viral tropical disease affecting both CNS and PNS. Hereby report a case of acute onset CIDP (A-CIDP) following primary infection with JE who presented as symmetric flaccid areflexic sensorimotor quadriparesis with subsequent clinical improvement with steroids and plasmapheresis.

Keywords: CIDP, JE, Neurotropic, Nerve conduction study

INTRODUCTION

Amongst neurological complications of JE virus, infectious polyradiculoneuritis is very rare. We report a case of relapsing typical CIDP presenting as an A-CIDP in its most recent clinical attack following primary infection with JE hypothesizing as post infectious disease. Although direct neurotropism and autoimmune response triggering by virus has been suggested, exact mechanisms by which JE involves both central and peripheral nervous system remains unclear till date.[1] CIDP is a heterogenous immune mediated demyelinating neuropathy with either a typical/ variant presentation, which can have mid-to long term neurological disability sequelae.

CASE REPORT

A 50 year old male with nil known comorbidities presented 2 weeks after a nonspecific febrile illness with a relapsing remitting illness in form of imbalance while walking with swaying to either side, aggravated by eye closure with cotton wool sensation while walking since 26 days, weakness of bilateral lower limbs in form of difficulty getting up from squatting position with buckling of both knees while walking since 26 days and tingling sensation of bilateral hands and feet since 26 days with no symptoms suggestive of cranial nerve involvement and with no dysautonomia and respiratory distress. No history of recent vaccinations but patient had recently travelled to his native place in Pondicherry (South India) in April, 20 days prior to onset of last clinical attack. Patient had history of similar complaints 9 years and 1 year back, which manged with indigenous medications with last episode leading to mild residual weakness of both lower limbs but not interfering with daily activities. On neurological exam, patient was conscious and oriented but had symmetrical flaccid weakness in bilateral upper and lower limbs with proximal more than distal weakness with upper limb proximal power MRC 4/5 and lower limb proximal power MRC 3/5, with areflexia of lower limbs, with decreased vibration and joint position sense in bilateral lower limbs up to knee with Rhomberg test positive and decreased touch and pain perception in bilateral lower limbs up to knee with normal sensory exam in upper limbs with normal cranial nerve, extrapyramidal and higher mental function examination. With clinical suspicion of CIDP, patient was further worked up with CSF study showing albumin-cytological dissociation with serum and CSF JEV-IgM (via ELISA) positive. Nerve conduction studies showed primary demyelination with secondary axonal changes consistent with EFNS-2021 criteria for typical CIDP. Blood sugars were normal,

retroviral status-negative, myeloma screening-negative, vasculitis panel-negative, CT scan of chest/ abdomen/ pelvis revealed no malignancy. MRI spine showed no compression of cervical and lumbo-sacral nerve roots.

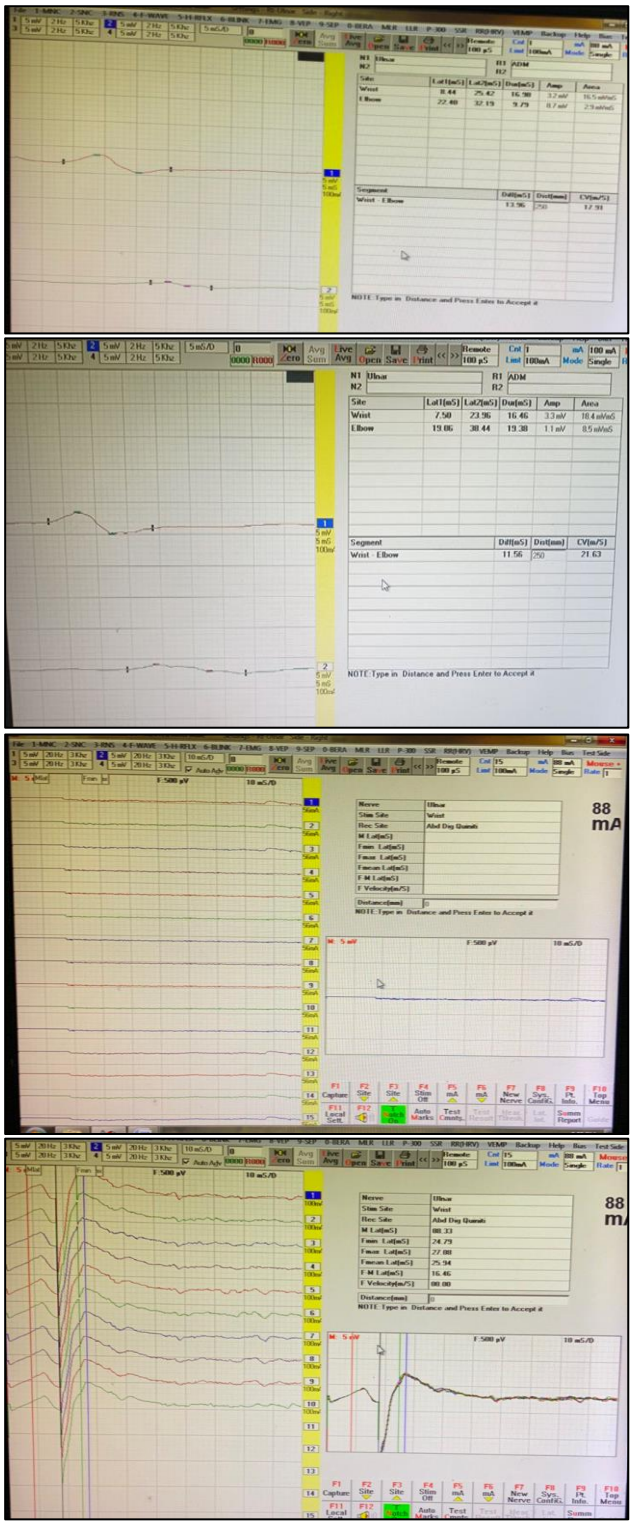

Figure 1: Nerve conduction study of bilateral ulnar nerves showing distal motor latency prolongation, compound muscle action potential amplitude reduction and reduced conduction velocities with F waves not obtained.

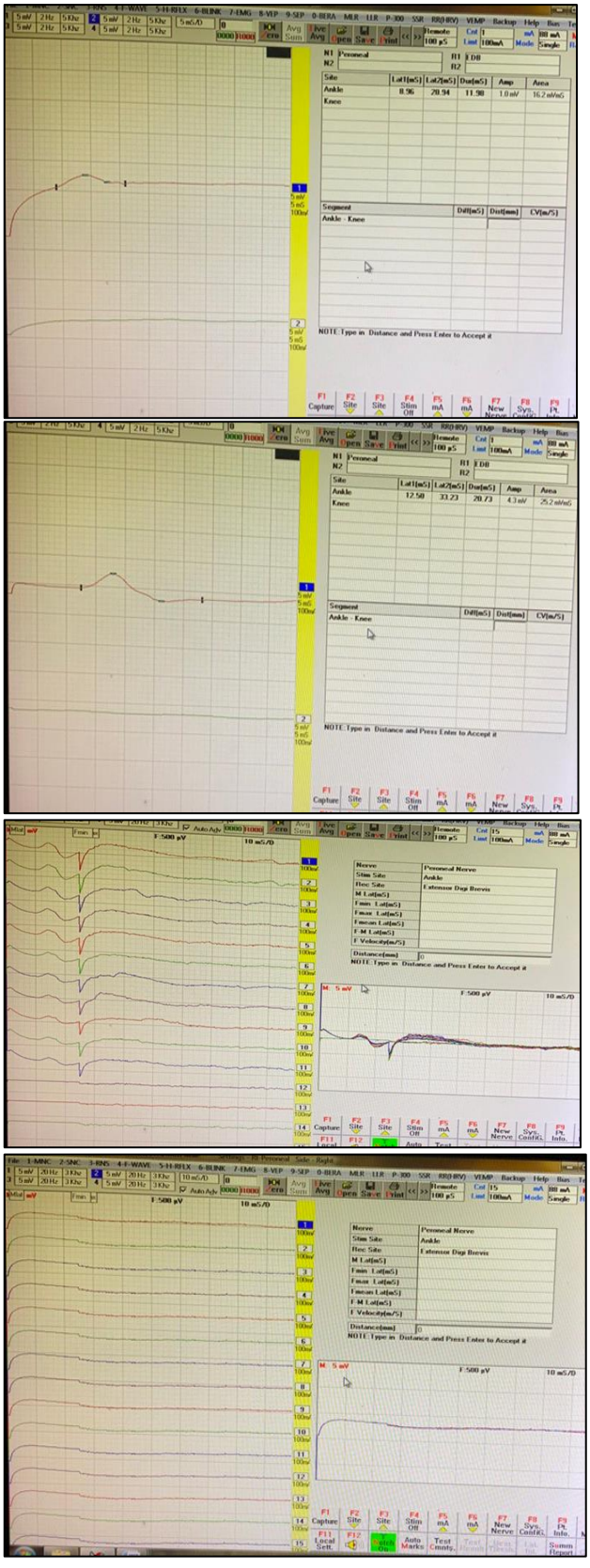

Figure 2: Nerve conduction study of bilateral peroneal nerves showing distal motor latency prolongation, compound muscle action potential amplitude reduction and reduced conduction velocities with F waves not obtained.

Patient initially received on day 4 of admission, IV methylprednisolone pulse therapy followed by oral steroids. On day 14 of admission, patient's neurological exam reviewed and since no clinical improvement in background of prior clinical attacks, he was started on plasmapheresis with 5 cycles on alternate days. Post 5 cycles of plasmapheresis, patient's motor power improved to MRC 4+/5 in both proximal upper and lower limbs with patient able to ambulate independently. He discharged on oral steroids and azathioprine. On follow up, evolution of disease was marked with complete recovery 6 weeks after onset of illness.

DISCUSSION

JE virus is a mosquito borne neurotropic flavivirus which is amongst the leading causes of viral encephalitis in the rural South East Asian and Indian subcontinent.[2] The most common neurological manifestation is that of an acute febrile encephalopathy with predominant extrapyramidal manifestations in the form of mask like facies, generalized hypertonia with cogwheel rigidity, tremor, choreoathetosis, opsoclonus myoclonus and rarely opisthotonos and rigidity spasms. The other reported neurological manifestation is that of a poliomyelitis like acute flaccid paralysis with anterior horn cell damage. In recent times post JE infection, a myriad of neurological manifestations including, immune mediated transverse myelitis, acute disseminated encephalomyelitis, NMDA receptor encephalitis and GBS like acute infectious polyradiculoneuritis have been reported. In endemic areas, JE mainly affects the children while the adult population in JE endemic areas are usually immune to JE through repeated asymptomatic exposures or vaccination.[3]

While a GBS like presentation has been associated post JE, CIDP occurrence post JE remains poorly characterized.[4] CIDP can present occasionally (13%) as an acute onset of CIDP, mimicking GBS. Similar to GBS, an infectious event can also trigger the onset of CIDP. However, unlike GBS, no infection has been consistently linked to onset of CIDP.

The diagnosis of JE was confirmed in our case by the demonstration of JEV-specific immunoglobulin M (IgM) in CSF by enzyme linked immunosorbent assay (ELISA) which is present in nearly all patients by day 7 of illness. Although JE typically presents as an acute infection with an initial febrile course lasting 2-3 weeks, sometimes the virus can persist in the nervous system leading to reactivation of latent JE. The association of JE with CIDP in our case could either be due to persistent viral infection sustaining the autoimmune response in CIDP or a recent antecedent viral infection triggering CIDP. Pathogenesis in this scenario could be explained by the JE virus inducing proliferation of myelin basic protein specific T lymphocytic cells leading to myelin destruction and demyelination.[5]

For the treatment of para infectious A-CIDP in our patient, there is no antiviral therapy with established efficacy in JE till date.[6] For JE-associated GBS, good response to treatment with corticosteroids and intravenous immunoglobulin has been described in literature, but use of plasmapheresis has not been reported. Regarding our case scenario, there was a paucity of clinical data on CIDP in background of JE infection, but nevertheless our patient responded to plasmapheresis.

CONCLUSION

The objective of this case report is to throw light on the occurrence of relapsing CIDP post JE that presented as an A-CIDP in the last clinical attack. Although JE typically presents as a monophasic illness, it can be considered in relapsing forms of neurological illness also. Our patient had significant functional recovery only after treatment with corticosteroids and plasmapheresis.

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