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Letter to Editor

Internal Medicine Section

Opsoclonus-Myoclonus Syndrome (OMS) Due to Dengue Meningoencephalitis

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Dear Sir,

A 60-year-old female presented to the department of Medicine with complaints of fever and headache since 5 days, rotatory irregular and rapid movements of both eyes and tongue since 4 days and two episodes of generalised tonic clonic seizures on the day of presentation. There was no history of cough, vomiting, abdominal pain, unconsciousness. On examination her GCS was E4V3M3, Pulse: 120 bpm, RR: 20/min, BP: 130/80 mmHg, Temperature: febrile to touch. Cardiovascular, respiratory system and abdominal examinations were normal. Patient was irritable but oriented. Higher functions, motor and sensory examinations were normal. There was visible Opsoclonous and myoclonus of tongue noted [Video-1].

Routine haemogram was normal. Absolute Platelet count was 80,000/cumm. Malaria card test was negative. Anti Leptospira Ig M was negative. Dengue IgM and NS1 were Positive. ELISA for HIV was negative. Liver function tests, Kidney function tests, & Thyroid profile, ANA, dsDNA were negative. Serum glucose, sodium, potassium, calcium were within normal limits. MRI Brain was normal. X ray chest, ultrasound of abdomen and pelvis was normal. Breast palpation was negative for any mass. CSF examination revealed TLC 280 cells with lymphocytic pleocytosis, Glucose-90 mg%, protein-140.1 mg%, CSF ADA was negative. CSF-PCR for HSV-DNA was negative. EEG in the inter-ictal period did not show any ictal activity. With the findings of fever, thrombocytopenia and positive Dengue serology, the final diagnosis Dengue Meningoencephalitis with Opsomyoclonus syndrome was done and patient was treated with Inj. Levetiracetam 500 mg iv BD, Analgesics and IV fluids. Her fever subsided in next 3 days, platelet count on 3rd day of treatment was 1,40,000/cumm. Opsomyoclonus (OMS) improved and the patient was discharged after 5 Days.

OMS also known as Kinsbourne syndrome is a rare neurological disorder that has a prevalence of 1 in 10,000,000 [1]. The mechanism behind OMS is auto-immune dysfunction of Purkinje cells in the dorsal vermis that leads to subsequent disinhibition of oculomotor fastigial region and damage of the omnipause cells in the pontine raphe nucleus [1]. It is characterised by high amplitude, arrhythmic, multidirectional, and conjugate ocular saccadic intrusions without intersaccadic latency causing oscillopsia [2]. It is also called saccadomania and associated with myoclonus of axial and/or appendicular musculature and ataxia [3]. It is frequently caused by neuroblastoma in children and breast or lung carcinoma in adults. Autoimmune diseases like Hashimoto encephalopathy and toxins are also known to cause OMS [1].

Dengue infection is most commonly encountered arboviral disease and the fastest spreading tropical illness in the world. Dengue may involve nervous system though it is considered a haematotropic virus. Out of the 4 serotypes, DENV1 and DENV2 are commonly associated with neurological manifestations. The index case was an adult with dengue infection who developed opsoclonus-myoclonus syndrome. The other common conditions associated with OMS in adult are Paraneoplastic manifestation, HIV infection, Herpes virus infection. A normal brain MRI, and negative serologic tests and CSF studies for the said infections were normal. There was no history that could point towards any malignancy. The patient had positive serological test for Dengue that established the diagnosis. OMS could be secondary to both dengue encephalitis and post-infectious immune injury [4].

The known neurologic manifestations of Dengue are encephalitis, meningitis, stroke, cerebellar syndrome, transverse myelitis, Acute Disseminated Encephalomyelitis (ADEM) [4]. OMS has been rarely encountered in dengue meningoencephalitis [5]. No specific anti-viral is available for the treatment of dengue encephalitis. The treatment is usually supportive in form of fluid resuscitation, thrombocytopenia management where indicated and anticonvulsant therapy were needed [4].

To conclude, OMS is a rare manifestation of dengue infection. Other possible causes of OMS in adults are paraneoplastic and infection with herpes simplex virus, HIV, mumps. The patient had a short history of fever, thrombocytopenia, a positive dengue serology and suggestive CSF findings to confirm the diagnosis. There are no evidence-based guidelines or consensus to treat dengue till date. Therapy is usually empirical and supportive. In present case, the fever subsided, platelet counts increased, and the OMS improved spontaneously with supportive treatment only.

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