Abstract

Chikungunya is a common tropical viral infection in India. The majority of patients have limited systemic manifestations. Neurological manifestations of chikungunya may be due to direct viral infection or immune mediated. We present a case of a 45-year-old male who presented with acute onset paraplegia with diminution of vision in the right eye. A detailed evaluation revealed a diagnosis of chikungunya myeloradiculitis with viral keratitis. The patient was treated with steroids followed by intravenous immunoglobulin and had a good recovery.

Keywords: Chikungunya, keratitis, myeloradiculitis

Introduction

Chikungunya is an arboviral disease transmitted by a mosquito bite (Aedes aegypti). The causative agent is the chikungunya virus which is a single-stranded RNA virus. Fever, polyarthralgia, and a maculopapular rash are hallmark manifestations of this disease. These manifestations are self-limiting and complete recovery occurs in most cases. Chronic and persistent arthralgia is a frequent complication in patients with severe chikungunya viral infection.31 Although rare, neurological manifestations can be the presenting feature of chikungunya infection and can leave long-term sequelae. We present a case of a 45-year-old male who presented with acute onset paraplegia with diminution of vision in the right eye and was diagnosed to have chikungunya myeloradiculitis with viral keratitis.

Case Report

A 45-year-old male presented with a history of fever, severe bilateral knee, and ankle joint pain and swelling for 3 weeks. After 10 days, he developed acute onset, severe, and continuous pain involving the lower back with radiation to the bilateral gluteal region. The pain was followed by symmetrical paraparesis which progressed over 2 days. This was associated with urinary retention and constipation along with loss of all sensory modalities below the level of the umbilicus. There was no history of recent vaccination. There was no history of any past medical illness. The patient also had excessive watering with pain and with blurred vision in the right eye for the past 2 weeks. This was associated with foreign body sensation and photophobia. There was no history of similar complaints in the left eye.

On neurological examination, the patient had hypotonia with a power of 0/5 in both lower limbs. There was a sensory level at D10 dermatome, absent deep tendon reflexes in lower limbs, and mute plantar responses. There was no involvement of the upper limbs. On local examination, mild bilateral knee and ankle joint pain and swelling were present.

On ocular examination, there was right eye corneal clouding. The right eye fundus could not be visualized, and in the left eye, fundus was normal. Pupils were bilaterally equal and reactive to light. Eye movements and other cranial nerve examinations were normal.

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were normal. Examinations of the cardiovascular, respiratory, and gastrointestinal systems were unremarkable.

On slit-lamp examination, there was a corneal epithelial defect with irregular shape and margins suggestive of a dendritic ulcer in the right eye [Figure 1]. Corneal sensations were absent in the right eye.

Magnetic resonance imaging (MRI) of the spine showed a T2 hyperintensity with corresponding T1 postcontrast enhancement extending from the T7-T11 cord level [Figure 2]. There was additional lumbar nerve root enhancement [Figure 3]. Thus, MRI was suggestive of thoracic myelitis with lumbar radiculitis.

All laboratory parameters including complete hemogram, renal and liver function test, thyroid function test, and blood sugar levels were normal. Neuromyelitis optica antibody, myelin oligodendrocyte glycoprotein antibody, oligoclonal band in both serum and cerebrospinal fluid (CSF), antinuclear auto antibodies-indirect fluorescent antibody, cytoplasmic antineutrophil cytoplasmic antibodies, and perinuclear anti-neutrophil cytoplasmic antibodies were negative. Serum angiotensin-converting enzyme levels were normal. Seromarkers including human immunodeficiency virus, hepatitis B surface antigen, and hepatitis C virus were negative. CSF study showed normal protein (52 mg/dl), normal glucose, and 10 cells (90% lymphocytic). Herpes simplex virus-polymerase chain reaction (PCR) and neurotrophic pan viral panel were negative. COVID-19 reverse transcription-PCR in nasopharyngeal swab and CSF was negative. High-resolution computed tomography of the thorax was normal. In view of the past history of fever with severe joint pain and swelling, we tested the patient for chikungunya. Immunoglobulin M (IgM) antibody by MAC ELISA for chikungunya was positive. On the basis of clinical presentation, laboratory, and radiological findings, a final diagnosis of chikungunya-associated thoracic myelitis with lumbar radiculitis with keratitis was made. The patient was initially treated with methylprednisolone 1 g intravenous for 5 days followed by a tapering dose of oral steroids for 4 weeks, but no significant improvement was seen. Therefore, after 1 week of intravenous methylprednisolone, intravenous Ig (2 g/kg) was given over 5 days. The patient showed significant improvement and was able to stand and walk with support within 14 days. In addition, the keratitis ulcer was treated with topical steroids. There was a good response over 14 days with no stain uptake and normalization of corneal sensation [Figure 4].

**DISCUSSION**

Chikungunya is an arboviral infection that has a self-limiting course; various neurological manifestations have been reported due to the neurotropic property of the virus. Along with neurological manifestations, the chikungunya virus is associated with complications of other systems, namely cardiovascular, renal, respiratory, hepatic, gastrointestinal, and adrenal systems.[2] Neurological disease following chikungunya

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**Figure 1**: Slit-lamp examination showing corneal epithelial defect with irregular shape and margins suggestive of a dendritic ulcer in the right eye

**Figure 2**: Gadolinium-enhanced T1-weighted magnetic resonance imaging of the spine (axial section) showing postcontrast enhancement (red arrows) at T7 cord level

**Figure 3**: Gadolinium-enhanced T1-weighted magnetic resonance imaging of the spine (axial section) showing postcontrast enhancement (red arrows) of the lumbar nerve roots
virus infection was first reported in 1964 in Madras, India.\textsuperscript{[3]} In a literature review of chikungunya-associated neurological disease of 856 cases, 796 (93.0\%) were in adults and children infected directly through mosquito, and 60 (7.0\%) were in neonates infected vertically from mother to child.\textsuperscript{[4]} The most common clinical presentation of neurological disease has been found to be encephalopathy followed by pure peripheral nervous system disorder.\textsuperscript{[4]}

The incidence of spinal cord disease after chikungunya infection is not exactly known, but it is likely to be less than that of encephalopathy. Most patients presenting with myelopathy, have more widespread central nervous system disease in the form of encephalopathy and peripheral nervous system disease (radiculopathy or neuropathy). Only a few patients have been reported with pure myelopathy syndrome.\textsuperscript{[4]} Spinal cord disease typically presents between day 1 and 3 weeks after the first clinical feature of infection.\textsuperscript{[4]} Chikungunya may affect the nervous system by direct viral neuroinvasion or by an immune-mediated response evidenced by upregulation of TLR2 gene (associated with innate immune response) in gray matter astrocytes.\textsuperscript{[3]} The occurrence of ocular manifestations in chikungunya fever also indicates severe disease and usually manifests from 4 to 12 weeks of acute disease.\textsuperscript{[4]} Patients with simultaneous occurrence of ocular disease with systemic manifestations suggest direct viral invasion presenting as conjunctivitis or anterior uveitis.

Late involvement of ocular tissue suggests a delayed immune response seen in cases of episcleritis, viral retinitis, panuveitis, and optic neuritis occurring secondary to antigenic mimicry. Corneal involvement in the form of viral keratitis is a relatively uncommon ocular manifestation of chikungunya.\textsuperscript{[7]} Our patient presented with myeloradiculitis with a keratitis ulcer which in itself is a rare presentation of chikungunya viral fever. The patient responded to immunomodulation suggesting an underlying immune-mediated response.

**Research quality and ethics statement**
We declare that the EQUATOR Network guidelines, notably the CARE guidelines were followed during the conduct of this report.

**Declaration of patient consent**
We certify that we have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his clinical information to be reported in the journal. The patient understands that his name and initials will not be published and due efforts will be made to conceal his identity, but anonymity cannot be guaranteed.

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Nil.

**Conflicts of interest**
There are no conflicts of interest.

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