IOSR Journal of Dental and Medical Sciences (IOSR-JDMS)
e-ISSN: 2279-0853, p-ISSN: 2279-0861.Volume 23, Issue 7 Ser. 2 (July. 2024), PP 01-03
www.iosrjournals.org

Opsoclonus Myoclonus Syndrome in Scrub Typhus with Dengue Positive Patient: A Rare Case Report

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Abstract:
Background: *Dancing eye syndrome or Opsoclonus Myoclonus Syndrome (OMS) is a distinct neurological entity. Febrile illness with OMS is a rare presentation with rapid, multi-directional eye movements (opsoclonus) and quick, involuntary muscle jerks (myoclonus).*
Methodology:
This rare case was seen in the Department of General Medicine, M.S. Ramaiah Medical College, Karnataka, India in December 2023.
Case details: *We present the case of a 22-year-old, young male, with no known comorbidities, hailing from Southern India, presenting with high-grade fever, headache and vomiting. During course of the hospital stay, the patient developed characteristic rapid, jerky eye movements that were multi-directional and sudden involuntary muscle jerks. Meningoencephalitis was suspected and he was initially treated for it, however neuroimaging and cerebrospinal fluid analysis was not contributory. Further workup revealed that he was Dengue IgM positive. Considering the epidemiological and clinical characteristics, Scrub Typhus titres were sent and it was positive, adequate treatment with doxycycline ensured complete resolution of his symptoms. This case report shows evidence to the rare presentations of infectious diseases around the world. OMS and encephalitis in a young patient could be debilitating if not aggressively managed and this case report reveals this importance.*
Cconlusion: *Early detection and management can improve clinical outcomes. To the best of our knowledge, this is the only reported case of dengue scrub typhus co-infection presenting as Opsoclonus Myoclonus syndrome.*
Keywords: *Opsoclonus Myoclonus (OMS), Dengue, Scrub Typhus, Encephalitis, Co-infections*

Date of Submission: 26-06-2024 Date of Acceptance: 04-07-2024

I. INTRODUCTION

Opsoclonus myoclonus syndrome (OMS) is a neuroinflammatory disorder, that is characterized by involuntary chaotic eye movements (opsoclonus) and sudden, jerk-like muscle contractions (myoclonus). Marcel Kinsbourne first described this syndrom in 1962, and it is also known by several other names including "dancing eyes–dancing feet," "Kinsbourne syndrome," "myoclonic encephalopathy," or "opsoclonic encephalopathy." OMS manifests with symptoms affecting vision, motor function, behavior, sleep, and language. It can be associated with underlying occult tumors, infections, or be idiopathic. Opsoclonus, a less common clinical finding, is frequently observed in opsoclonus myoclonus ataxia syndrome (OMAS), which can be paraneoplastic or postinfectious. Opsoclonus with or without OMAS has been reported in association with various infections, highlighting infection-associated opsoclonus/OMAS (IAO) as a distinct entity requiring brief immunosuppression and symptomatic management and typically having a favorable prognosis.[1]

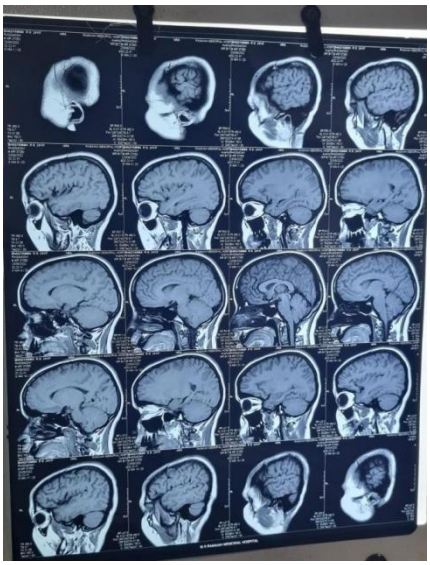
Scrub typhus, caused by Orientia tsutsugamushi and transmitted by the bite of the larval form of trombiculid mites, is prevalent in the "tsutsugamushi triangle" from Pakistan and Afghanistan to northern Japan and northern Australia. Patients usually present with a short febrile illness with a distinctive eschar, scrub typhus is recognized to involve neurological manifestations like seizures, aseptic meningitis, and uncommonly, opsoclonus.[2-3] Dengue is a mosquito-transmitted virus, and dengue fever is the leading cause of arthropod-borne viral disease worldwide, posing a significant global health concern. Dengue is recognized for causing neurological complications such as meningoencephalitis, transverse myelitis, and Guillain–Barre syndrome (GBS), its association with OMS is very rare.[4-9]

Previously only few cases of dancing eye syndrome were reported associated with Scrub typhus and Dengue individually.[6-8] Here, we present a case of OMS associated with concurrent dengue and scrub typhus infection.

II. CASE DETAILS

A 22-year-old gentleman, who was a student of engineering and no prior illnesses, presented to the emergency room with a history of high-grade fever with chills since a week. He gave history of holo cranial, persistent headache and vomiting for the last five days. Though the patient had no history of prior health concerns, he gave the history of pesticide spraying in rice fields about a fortnight before developing his symptoms. The patient visited a primary health care center with the above complaints, where he was administered antipyretics and intravenous fluids, however, he persisted in having high-grade fever spikes and multiple episodes of vomiting. At arrival, his vitals were stable and he was conscious, with no focal neurological deficits or meningeal signs. Had a soft, nontender, nondistended abdomen with warm, well-perfused extremities. The patient was admitted with the suspicion of meningoencephalitis and promptly started on antibiotics- intravenous third-generation cephalosporin *Ceftriaxone*, intravenous *Acyclovir* anti-cerebral edema measures, anti-emetics, and supportive therapy. Initial laboratory workup of plasma and serum- renal, liver function testing, complete blood picture- yielded no new clues to his condition. About 2 hours into the course of hospital stay, patient developed rapid, jerky eye movements that were multi-directional and when he sat upright, he developed sudden involuntary muscle jerks, the characteristic opsoclonus-myoclonus. Fundoscopy revealed grade 2 papilledema, revealing raised intracranial pressure. A guarded lumbar puncture was performed, CSF analysis- 5 cells, 100% Lymphocytes. CSF was also processed for CBNAAT for Tuberculosis and HSV PCR-negative. The patient persisted in having fever spikes, requiring further investigation. *Dengue Serology was sent suspecting Dengue Encephalitis- IgM-ELISA- Positive with significant titers.* Cultures from CSF and blood had no growth. Neuroimaging was within normal limits. Despite conservative management for Dengue encephalitis, and the patient continued to have fever spikes and myoclonic jerks and opsoclonus.

Keeping the relevant history of his exposure to shrubbery and rice fields, Scrub Typhus IgM Elisa was sent, which came back positive with significantly high titers. Patient was started on intravenous *Doxycycline.* On Day 2 of initiation of the new therapeutic regimen, the patient had no further fever spikes. He was discharged with a course of oral antibiotics- cephalosporin and doxyxycline. Opsoclonus-Myoclonus resolved over 2 weeks of complete treatment.



MRI with Contrast -Brain- Normal Brain and Meningeal Parenchyma

III.DISCUSSION

In the current study, we reported a rare case of OMS among 22 year old male with scrub typhus and dengue.

Previously **Garg et al-** reported a case with OMS as an initial manifestation of scrub typhus in a 23-year-old pregnant female, who presented to the emergency room with severe fever for one week, She showed abnormal movements of her eyes and limbs from day two of fever onset. [9]

Desai et al reported OMS in a 14-year-old patient who presented with fever, confusion. On examination, the patient had opsoclonus in his eyes along with cortical myoclonus in his hands and body. He showed low platelets, normal brain imaging. CSF analysis showed lymphocytic pleocytosis. He was treated conservatively and had spontaneous improvement by 5th day and complete recovery in 2 weeks. [10]

Sani et al reported opsoclonus in the setting of an acute infection/febrile illness among 15 children. Myoclonus was seen among 2 children, ataxia was seen among 4 and behavioral abnormalities were seen among 4 children.

All these patients had an associated neurologic or nonneurologic illness- scrub typhus or tuberculous meningitis (n or encephalitis or acute cerebellitis or clerosing panencephalitis.[11]

Our case highlights the increasingly variable and complex neurological presentation of scrub typhus with Dengue. OMS is a well-characterized paraneoplastic syndrome with a likely antibody-mediated mechanism. Parainfectious OMS, unlike its paraneoplastic counterpart, seems to have a good prognosis when the underlying infection is treated.

IV. CONCLUSION

Dengue Virus, belonging to *Flaviviridae* has a myriad of presentations, despite being a hematotropic virus. Scrub typhus is caused by *Orienta tsusugamushi*, chiggers being the vectors. Co infection with the infectious agents lead to an aggressive, debilitating neurological illness in this young individual. Early detection and management can improve clinical outcomes. To the best of our knowledge, this is the only reported case of dengue scrub typhus co-infection presenting as Opsoclonus Myoclonus syndrome.

There were no conflicts of interest

Ethicals aspects: Informed consent was taken from the patient who participated in the study.

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