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LETTER TO THE EDITOR

Scrub typhus presenting as diaphragmatic myoclonus

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Sir, Van Leeuwenhoek's disease, commonly known as diaphragmatic myoclonus, diaphragmatic flutter, moving umbilicus syndrome, or dancing-belly syndrome, is a high-frequency, involuntary, non-suppressible, distressful contraction of diaphragmatic muscles [1]. It has been associated with several cardiopulmonary intervention procedures, cardiac arrhythmias, stroke, encephalitis, demyelination, lesions involving the cervical spine, irritation of a phrenic nerve by cardiac electrode dislocation, metabolic abnormalities, and SARS-CoV-2 infection [1–3].

Scrub typhus is an acute febrile infectious disease common in the "Tsutsugamushi triangle," caused by Orientia tsutsugamushi, a mite-borne rickettsial zoonosis [4–6]. Meningitis, encephalitis, cranial nerve paresis, transverse myelitis, and polyneuropathy are the most well-documented neurological manifestations [4–6]. By contrast, diaphragmatic myoclonus has not been reported as a presenting manifestation of scrub typhus. We report the case of a healthy man who presented with a recent onset diaphragmatic myoclonus and fever but without the classic dermatologic manifestation ("eschar"). After excluding common infectious, autoimmune, and neoplastic causes, he was finally diagnosed with scrub typhus associated with diaphragmatic myoclonus, which responded to doxycycline and azithromycin.

A previously healthy 45-year-old man from rural West Bengal (India) was admitted to the emergency room with complaints of fever and headache for the last 5 days and abrupt onset abnormal "involuntary and jerky" movements simultaneously involving shoulder girdles and abdomen for the last 2 days. He did not have any family history of neuropsychiatric disorders, history of addictions, and long-term intake of any medications. His vital signs were normal except for a temperature of 101.1ºF and a pulse of 116 bpm. Pulmonary and cardiac auscultation and abdominal examination were also unremarkable. The neurologic examination revealed normal higher mental functions. He had no opsoclonus or other abnormalities in the cranial nerves. He showed frequent, involuntary, rapid, brief, arrhythmic jerks suggestive of subcortical myoclonus at rest (which increased with auditory stimuli) involving the shoulder girdles. There were also abnormal, involuntary, arrhythmic, inward, and outward movements of the abdominal wall suggestive of diaphragmatic myoclonus [Video 1]. Action myoclonus and startle responses were also present. No abnormal movements were noted involving the pelvic girdle and lower limbs. No abnormal orolingual movements were observed.

Blood analyses revealed neutrophilic leucocytosis, raised erythrocyte sedimentation rate, C-reactive protein levels, and mild transaminitis. Blood glucose profile, thyroid function, and renal function tests were otherwise normal. Autoimmune and paraneoplastic encephalitis profile including anti-thyroid antibodies was negative. Magnetic resonance imaging of the brain and the spinal cord and electroencephalogram were

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normal. Cerebrospinal fluid (CSF) study revealed increased protein (160 mg/dL) and lymphocytic pleocytosis (67 cells, all lymphocytes) and lower glucose levels (30 mg/dL). Relevant neuroviruses tested by a polymerase chain reaction (PCR) were negative, just like tests for neurotuberculosis, neurosyphilis, neuroborreliosis, and subacute sclerosing panencephalitis. Malaria and dengue were also ruled out. Paired sera (CSF and serum) for scrub typhus PCR were positive. He was put on 200 mg/day oral doxycycline and azithromycin 600 mg/day. After 72 h of antibiotic therapy, he became afebrile, and his headache had reduced. After five days of antibiotic therapy, the myoclonic jerks started to disappear and completely disappeared after eight days.

Diaphragmatic myoclonus is an exceptional disorder characterized by high-frequency, involuntary contractions of diaphragmatic muscles, with semirhythmic jerking and displacement of the abdominal wall, frequently along with contraction accessory respiratory muscles [1, 2]. Diagnosis of diaphragmatic myoclonus as presenting manifestation of scrub typhus requires a high index of suspicion due to its rarity and varied presentations such as hiccup, epigastric pulsation, palpitation, and hyperventilation, mostly to non-neurologists [1, 2]. Atypical manifestations of scrub typhus are on the rising trend in the tropics, making it more difficult to diagnose this multi-systemic infection. In this sense, our patient did not have the pathognomonic "eschar" on his body, similar to other reported cases [6].

Treatment of diaphragmatic myoclonus comprises, besides treating the offending cause, oral pharmacotherapy (phenytoin, carbamazepine, and haloperidol, among others), phrenic nerve crushing, and botulinum toxin administration[1,2]. In our case, antibiotic therapy for scrub typhus infection resulted in decreased abnormal movements, with following resolution *ad integrum* after eight days.

In closing, in all cases of acute onset movement disorders associated with febrile illness in tropics/subtropics, scrub typhus infection should be included as a differential diagnosis, although neuroimaging is unremarkable, and there is no eschar. Delay in diagnosis may lead to multi-organ-dysfunction syndrome and death in this otherwise curable disease.

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Declarations

Ethics statement Informed written consent was obtained from the patient involved in this study.

Competing interest The authors declare no competing interests.

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