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Case Report

Cranial polyneuropathy and triparesis due to mononeuritis multiplex in a case of scrub typhus

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ABSTRACT

Scrub typhus with neurological manifestations are often described. We present here unusual neurological manifestations in a patient with Scrub typhus. A 49 years old male presented with continued fever for the last 12 days along with headache and myalgia. He had left sided oculomotor nerve palsy along with palatal palsy and triparesis (except left upper limb) of lower motor neurone type. Electro-diagnostic studies showed asymmetric axonal sensorimotor neuropathies of multiple nerves in paretic limbs, suggestive of mononeuritis multiplex. Scrub typhus was diagnosed by the presence of IgM antibody in serum. He responded well with doxycycline therapy. This type of neurological presentation is extremely rare in scrub typhus and possibly has not been described earlier to the best of our knowledge.

Keywords: Scrub typhus, Cranial polyneuropathy, Triparesis, Mononeuritis multiplex

INTRODUCTION

Scrub typhus is acute febrile illness, caused by orientia tsutsugamushi and is transmitted to humans by trombiculid mites. It is commonly seen during monsoon and post monsoon season in our country, causing lot of morbidities and occasional mortality. It involves all organ system of body commonly characterized by fever, rash, lymphadenopathy, myocarditis and pneumonitis. The most characteristic skin manifestation is Eschar which is seen in about 4-46% in Indian population.[1,2] Neurological involvements, both peripheral and central nervous system are well known.[3] Most common CNS manifestation is meningo-encephalitis; also seen are cerebral infarction, cerebral venous sinus thrombosis, ADEM and cranial neuropathies (mostly isolated).[4] PNS involvement with GBS, mononeuritis multiplex, Brachial plexopathy and peripheral plexopathies has been described. Here we describe an unusual neurological manifestation of Scrub typhus with multiple cranial nerve palsies and triparesis (involving both lower limbs & right upper limb) due to mononeuritis multiplex.

CASE REPORT

A 49 years old male presented to us with history of continued fever for the last 12 days. Five days after the onset of fever, he suddenly developed drooping of left upper eyelid with visual disturbances when tried to look with both eyes. After a day, he felt weakness of left lower limb, more distally. After about one week, he developed weakness of right lower limb and then right upper limb but his left upper limb remained normal (triparesis). Occasional paraesthesia of involved limbs were complained of. No history of sphincter disturbances, headache, vomiting or LOC were seen. Initially he consulted a local hospital and was then transferred to our hospital. We also noticed his nasal intonation of voice then and on enquiry he complained of occasional dysphagia.

Examination revealed a febrile patient (temperature= 102.4 degrees of Fahrenheit) with normal sensorium, mild pallor and was dehydrated. He was hemodynamically stable. No skin rash, eschar or lymphadenopathy was found. Other systemic examinations were non-contributory. Neurological examination showed left sided oculomotor nerve palsy (pupil sparing) with palatal palsy (right>left) along with loss of gag reflex. Fundus examination was normal. His visual disturbances were likely due to diplopia from 3rd nerve palsy. Motor system examination showed weakness of lower limbs (left>right), distal> proximal. Right upper limb was also weak (distal>proximal). Left upper limb power was normal. Affected limbs had hypotonia and hyporeflexia. Plantar response was flexor.

No objective sensory loss could be demonstrated. No neck stiffness was found. He was non ambulatory owing to weakness of both lower limbs. So, the patient had multiple cranial neuropathies characterised by peripheral type of left oculomotor, glossopharyngeal and vagus nerves. He had lower motor neurone type of triparesis, evidenced by asymmetric both lower limb paresis and right upper limb paresis (distal>proximal). The cause of fever was undetermined at that moment.

Investigations revealed peripheral lymphocytosis and impaired LFT with altered A:G ratio and transaminitis. Associated hyponatremia was noted (Na+=118 mEq/L). Blood and urine cultures were negative. Other common infections like typhoid, malaria, dengue, leptospira were excluded. Chest X-ray and USG of whole abdomen were non-contributory. Electrophysiological studies showed multiple motor and sensory nerve involvement in all three involved limbs, mainly axonal type suggestive of mononeuritis multiplex. MRI (plain+contrast) of Brain and whole spine was done which came out to be normal. CSF study was done which showed cell count of 10 (all lymphocytes) with normal glucose and increased protein level of 140 mg/dl. CSF ADA was normal. India ink preparation was negative.

To find the cause of fever, scrub typhus IgM was sent which came positive. The patient was put on doxycycline (initially IV for one week, then orally). After 2 days he became afebrile. After 5 days of therapy there were signs of neurological improvement- partial improvement of ptosis with improvement of nasal intonation and dysphagia.

Limb weakness partially improved during hospital stay. He was discharged on 10th day with oral doxycycline therapy. At the time of discharge his ptosis further improved, lower cranial nerve palsy corrected and he could stand with support. Physiotherapy was advised.

Follow up after 3 weeks in OPD showed total improvement of Oculomotor nerve palsy and he was ambulatory with support.

DISCUSSION

O. tsutsugamushi, the causative organism of scrub typhus is an obligate intracellular parasite of various phagocytes that invade nervous system as a part of systemic infection. Scrub typhus was diagnosed in our patient by the presence of IgM antibody in serum.[1] Neurological involvement in scrub typhus has been well documented in literature.[3] Meningitis/meningo-encephalitis is the most common manifestation.[4] But our patient did not have meningo-encephalitis both clinically and investigation wise. His brain imaging was normal. Cranial nerve palsy in Scrub typhus has been described in literature.[5,6]

Limb weakness of GBS type in 5 patients out of 134 patients has been described by Kularatne et al.7 GBS like limb weakness are seen in literature.[8,15]

There are reports of Brachial plexopathy, pyramidal syndrome, deafness and otalgia.[9,11]

Multiple cranial nerve palsy with meningitis and cerebellitis is also described.[13] Limb weakness of GBS type in 5 patients out of 134 patients has been described by Kularatne et al.[7] GBS like limb weakness are seen in literature.[8,15] Meningo-encephalitis followed by GBS has been documented by Atul Phillips et al.[16] Similarly, Kim et al described a patient of polyneuropathy and cerebral infarction.[17] There are reports of Brachial plexopathy, pyramidal syndrome, deafness and otalgia.[9,11] But our patient did not have features of GBS as he had asymmetric lower limb weakness (distal>proximal) and only right upper limb weakness (Triparesis). This type of weakness can be explained by mononeuritis multiplex. This was also evidenced from electrophysiological studies. This type of presentation is very rare in Scrub typhus. Heyakawa et al described a patient of Scrub typhus with mononeuritis multiplex, acalculous cholecystitis and aseptic meningitis.[12]

Neurological manifestations of our patient were very atypical due to simultaneous involvement of multiple cranial nerves and triparesis due to mononeuritis multiplex which responded with specific therapy. The probable mechanism of such involvement is direct invasion of the organism in the nervous system causing acute stage vasculitis or secondary immune mechanisms causing vasculitis of vasa nervosa of nerves. Further studies with nerve biopsies will possibly be more informative (not done in our patient).

CONCLUSION

Scrub typhus can present with variety of neurological manifestations. Presentations with simultaneous multiple cranial nerves and triparesis due to mononeuritis multiplex is extremely rare and has not been described in literature. We should keep our minds open so that proper diagnosis and management can be given in due time with satisfactory improvement.

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