

Case Report

Scrub Typhus with Unilateral Parotitis and Encephalopathy : A Rare Case Report and Literature Review

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Scrub Typhus is an acute febrile illness recently re-emerging. Scrub Typhus is a zoonosis caused by *Orientia tsutsugamushi* (formerly *Rickettsia*). Though in majority of the cases Scrub Typhus has self limiting course, it may involve any organ resulting in multiorgan dysfunction. Scrub Typhus has to be thought of in the differential diagnosis of febrile illness. Here, we report a case of Scrub Typhus presenting as unilateral parotitis and multiorgan dysfunction. Doxycycline administration rapidly altered the clinical course. Knowledge about the non-specific clinical presentations and complications of Scrub Typhus, will help in early recognition and treatment so that complications can be avoided.

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Key words : Scrub Typhus, Parotitis, Multiorgan Dysfunction.

Scrub typhus is caused by *Orientia tsutsugamushi*, a mite born obligate, intracellular gram-negative coccobacilli, which is widely distributed throughout the South Pacific Asia¹. The original name of Scrub Typhus, that was given by Hashimoto in 1810, is "tsutsugamushi disease." It is transmitted by the bite of infected larvae of trombiculid mite (Chiggers). Primary vector for the disease is *Leptotrombidium deliense*. Humans are infected when they come in contact with infected mite. The hot and humid climate and tropical weather are favorable conditions for mite activity and disease transmission. Incubation period is 6-21 days².

In India initial cases were isolated during the socio-political turbulent times of 1930s³ and a large number of cases were initially described during World War II among the troops posted in the hilly regions of North East India⁴. The first outbreak of Scrub Typhus in India was reported in 2003-2004, when a cluster of paediatric patients presented with fever, hepatosplenomegaly and eschar. Scrub Typhus accounts for 25.3% of acute undifferentiated febrile illness in India with a community sero-prevalence of 34.2%⁵.

Scrub Typhus is an acute febrile illness with a wide range of clinical presentation starting from self-limiting illness with rash (50%), eschar (40%), lymphadenopathy, fever, headache, myalgia to life threatening complications including encephalitis⁶, interstitial pneumonia, ARDS, acute renal failure and acute hepatic failure. This possesses a great diagnostic challenge and dilemma while approaching an ill febrile patient. It is important to differentiate Scrub typhus from dengue, leptospirosis, malaria and enteric fever etc, so that appropriate treatment can be initiated.

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Editor's Comment :

- The first clue to diagnose a case of scrub typhus is high index of suspicion in primary care physician particularly in this subcontinental scenario. Every case of Fever of Unknown Origin (FUO) should be searched for Scrub typhus due to its wide spectrum of presentation. Hence diagnosing and treating early can easily prevent potentially lethal complications.

The main pathogenesis is either focal or disseminated vasculitis or perivasculitis that may damage organ such as the kidney, lung, liver, brain and skin⁷. Late diagnosis of Scrub typhus and inadequate treatment may lead to complications with multiorgan involvement. Though the median mortality rate of untreated case is 6%, that of treated case is 1.4%, the mortality rate may increase up to 70% without adequate treatment^{8,9}.

Despite a few studies on this topic from this region, it is still being grossly under diagnosed due to its nonspecific clinical presentation, limited public awareness, low index of suspicion among the clinicians and lack of diagnostic facilities in some places.

CASE REPORT

A 25-years-old, male, occasional alcoholic for 2 years, was admitted in Midnapore Medical College & Hospital, West Bengal with yellowish discolouration of eyes and urine for 14 days. He also complained of high grade fever for 10 days, right sided progressive painful facial swelling with difficulty in chewing and swallowing for 7 days and altered sensorium for 2 days. On admission he was febrile, dyspnoeic, pulse - 100/min, MAP - 58 mm of Hg, dry tongue with CRT >3 sec, jaundice +, SPO2 92% in room air and RBS - 85 mg/dl. One black eschar (1x1cm) was seen over the forehead and right parotid gland was swollen (Fig 2). On palpation, RUQ of abdomen exhibited tenderness without hepato- spleenomegaly. On auscultation, S1 and S2 was audible, and vesicular breath sound in both the hemithorax was decreased (Right > Left). The patient was drowsy with a GCS of 10/15(E3V3M4), bilateral flexor

plantar response and bilaterally reactive pupil without neck rigidity.

On admission, relevant investigations showed: TLC- 22,600/mm³ (N: 86, L:8, M:2, E:2), Haemoglobin - 9.5mg/dl, Platelet - 1,73,000/mm³, ESR - 62 millimetres/ hour, CRP - 41.53 mg/dL, microcytic hypochromic anaemia and toxic granules in neutrophils on peripheral blood smear. Thick and thin smear for MP/MPDA was found to be negative. Biochemical parameters revealed: serum urea - 220 mg/dl, creatinine - 2.69 mg/dL, serum Na⁺ - 132 mmol/L, serum K⁺ - 4.22 mmol/L, total bilirubin 35.79 mg/dl with conjugated bilirubin 26.03 mg/dL, ALT/AST 482/316 IU/L, ALP 382 IU/L, serum albumin 2.6 g/dl, serum amylase/Lipase 132/78 IU/L, PT 22.32 sec and INR 2.4. Patient was found to be non-reactive for HIV I, II, HBsAg, Anti-HCV RNA, Hepatitis A IgM, Leptospira IgM, Dengue IgM and NS 1ag. After 48-hour, 3 sites blood (both aerobic and aeorobic), and urine culture and sensitivity showed no growth. Sputum for AFB/CBNAAT and subsequently TB Quantiferron Gold was found to be negative. USG whole abdomen showed mild hepatomegaly, grade I fatty infiltration and increased echogenicity of Kidney (B/L). Chest X-Ray AP in supine position showed right sided minimal pleural effusion. Scrub IgM antibody with ELISA method was found to be positive, which was confirmed by Immunofluorescence method.

Patient was shifted to CCU, where he was conservatively managed with IV fluid, vasopressors, broad-spectrum antibiotics, 6-units of FFP and subsequently 2 settings of bedside haemodialysis. After diagnosis of Scrub typhus was made, patient was put on injection IV Doxycycline 100 mg BD.

2D Echo with M mode was done to rule out vegetations. CSF study revealed lymphocytic predominance with 12 WBCs, sugar - 52 mg/dL and protein - 75mg/dl, with a negative Gram Stain and Z-N Stain. CT scan of the neck and face showed the features of Parotitis (Fig 1). FNAC was suggestive of necrotizing Parotitis.

After a 10 days course of IV doxycycline, the patient showed dramatic clinical improvements. He became afebrile, swelling decreased (Fig 2), mentation and appetite improved. The laboratory parameters before and after the treatment are enlisted below Table 1.

DISCUSSION

Scrub Typhus affects one million people around annually. Trombiculid mite (Chiggers) of the Leptotrombidium genus, which feeds on wild rats, infects human accidentally who are living in Sub Himalayan, dense forest area or the people whose livelihood depend



Fig 1 — CECT of neck and face showing the features of Parotitis



Fig 2 — Disappearance of Parotid Swelling after 10 days of Doxycycline therapy: right (before), l

mainly forest and agriculture². The rickettsial diseases which were once thought to have disappeared from India, are re-emerging as zoonosis in several parts of India such as Kashmir, Himachal Pradesh, Sikkim, Darjeeling (West

Table 1 — Comparison of the laboratory parameters before and after Doxycycline therapy

Before	Parameters	After
22,600	TLC (/mm ³)	4,300
41.53	CRP (mg/dl)	3.56
220	Urea (mg/dl)	16
2.39	Creatinine (mg/dl)	0.62
35.79	Bilirubin (mg/dl) (total)	4.25
26.03	Bilirubin (mg/dl) (conjugate)	3.15
316	AST (IU/L)	56
482	ALT (IU/L)	78

Bengal) and Western Ghat. Aetiological factors once thought eradicated have re-emerged due to migration of labour from other states, poor hygiene in public places and improper waste disposal.

The overwhelming clinical manifestations of Scrub Typhus are attributed to systemic vasculitis and endothelial dysfunction¹⁰. Signs and symptoms of Scrub Typhus are nonspecific and clinical features may mimic other febrile illness. Though most of the cases are self limiting in nature with spontaneous recovery, delay in diagnosis and inadequate treatment may lead to complications including Multi Organ Dysfunction (MOD). Though eschar is the most useful diagnostic clue of Scrub Typhus, many Indian studies revealed eschar in less number of patients¹¹.

Early use of antibiotics can prevent life-threatening scrub typhus. The effective antibiotics against Scrub Typhus are doxycycline, chloramphenicol, tetracycline and azithromycin. Azithromycin may be more tolerable than doxycycline with same effect. The clinicians should avoid rifampicin as a first-line agent due to the low-certainty evidence and the risk of resistance in undiagnosed tuberculosis¹².

Similar presentation of Scrub Typhus had been previously reported from Kasturba Medical College, Manipal in 2017, where a 38 year old male presented with fever, altered sensorium, parotid swelling and MODS¹. AKI was reported in 13-27% of patients in two different Indian studies. Parotitis is rare clinical manifestation of Scrub Typhus with MODS. Only a very few literature is available to support its rampant in southern districts of West Bengal¹³. This case report will help the clinicians to get insight about the diversity of this potentially fatal condition and increase the index of suspicion while dealing with an acute febrile patient with MOD.

CONCLUSION

Rickettsial diseases are prevalent but remain underdiagnosed in India. Scrub Typhus should be differentiated from other causes of febrile illnesses of short duration because of similar clinical and laboratory features. When patients present with fever and multi organ involvement, Scrub Typhus should always be considered in the differential diagnosis. High index of suspicion and prompt initiation of treatment will reduce the morbidity and mortality.

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