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Case Report

Internal Medicine Section

Trichinellosis: A Case of Life-threatening Myositis and Myocarditis in Garhwal Hill Region

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ABSTRACT

Human Trichinellosis is now a rare zoonotic disease. Although the disease is not uncommon, rather non specific symptoms make the disease, a rare thought differential in acute febrile gastroenteritis. Incidence has decreased owing to healthy eating habits. The infection usually manifests with subclinical disease presentation, and acute gastroenteritis; however occasionally leading to life-threatening myocarditis, myositis and seizure. Diagnosis requires a low threshold for suspicion and evaluation. Muscle biopsy serves as the definitive diagnostic method. Most often it resolves without any treatment whereas severe cases require high dose intravenous glucocorticoid and antihelminthic. Here, authors report a case of a 23-year-old female patient, who presented with life-threatening acute onset, progressive pure motor quadriparesis with myocarditis. After detailed clinical and laboratory investigations, the possibility of acute infectious aetiology with eosinophilia, myocarditis, and myositis was diagnosed, and a muscle biopsy was performed. The patient was diagnosed with Trichinellosis after ruling out other causes of acute motor paralysis. The patient improved after albendazole and glucocorticoid therapy. The case represents an uncommon cause of myositis and myocarditis. Detailed review of the history and epidemiology of the infectious disease should always be given priority for early diagnosis. The case also gives the importance of timely treatment which was life-saving.

Keywords: Cardiogenic shock, Creatine kinase, Quadriparesis, *Trichinella spiralis*, Zoonotic disease

CASE REPORT

A 23-year-old female patient from Tehri Garhwal region of Uttarakhand, India, presented to the Emergency Department in our hospital with fever, body pain and generalised weakness for five days with no prior co-morbidity. She was apparently asymptomatic before she developed high grade, undocumented, intermittent fever for five days with no diurnal variation and was relieved on taking symptomatic treatment. It was associated with diffuse myalgia and generalised body weakness. The weakness started in all four limbs simultaneously and progressed in severity over four days and became so debilitating for the patient that she was unable to stand and walk without support. The patient had difficulty getting up from sitting and squatting positions, so she was using a diaper for defecation and urination by fifth day. She also had difficulty in combing her hair, taking food on her own.

However, there was no stiffness in her legs, slipping of slippers while walking, holding objects in hand. She was able to perceive pain and temperature, and had normal bowel and bladder habit. The patient's condition deteriorated further and by fifth day, she became bed-bound. She did not give a history of any snake or insect bite. On the same day, she complained of retrosternal chest pain, which was sharp, pleuritic in nature, lasted for one day, not associated with syncope, sweating, radiation to upper limb or jaw, associated with palpitation. It was associated with bilateral leg swelling, more in evening and associated with fullness around eyelids and facial puffiness. There was no history of loose motions, vomiting, sore throat, cough, rashes, seizure, altered mentation, pain abdomen, and bleeding from any site or decrease in urine output.

Patient also did not gave the history of night sweats, oral ulceration, hair loss, joint pain and swelling, photosensitivity before this episode. There was no history of any chronic corticosteroid use. Asking a leading question, she gave a history of acute gastroenteritis 15 days back after consuming 'PRASAD' in a community feast which she admitted to contain pork, and had developed seven to eight episodes of watery diarrhoea associated with vomiting and fever which subsided in two to three days after taking over-the-counter medication. There were no similar complaints in the past as well as in the family. Patient presented on fifth day of illness to our hospital.

On examination, she had tachycardia of 120 beats/minute, and tachypnoea of 20 beats/minute with tenderness and mild local rise in temperature in all four limbs. She had bilateral pitting type pedal oedema and eyelid swelling with no evidence of icterus, cyanosis, clubbing, and lymphadenopathy.

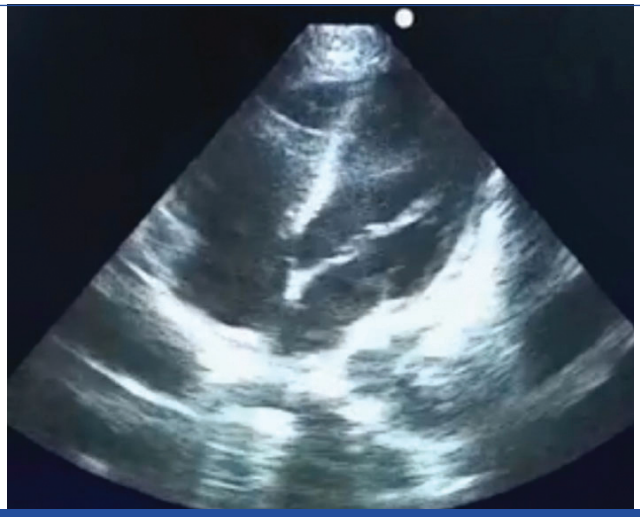
Her single breath count was significantly decreased, initially till 16. Systemic examination revealed normal tone, decreased power 2/5 in all four limbs, and decreased reflexes with plantar flexor response. Based on history and physical examination, an initial syndromic diagnosis of acute febrile illness with pure motor quadriparesis with an antecedent history of acute gastroenteritis was made. Differential diagnoses of infectious myositis, and immune mediated inflammatory myositis, Guillain-Barré were made. She was evaluated and the investigations are presented in [Table/Fig-1].

Investigations	Normal value	At admission (Day 1)	After treatment (Day 12)
Haemoglobin (g/dL)	11.5-15	10	10.5
Total leucocyte count (/mm^3)	4-10×10^9	7.940	4.94
Eosinophil count (%)	1-6%	10% (day 1) →7% (day 3)	3%
Platelet count (lakh/mm^3)	1.5-4.5	3.67	5.74
Total bilirubin (mg/dL)	0.3-1.2	1.12	0.67
Direct bilirubin (mg/dL)	<0.20	0.53	0.12
Serum Glutamic Pyruvic Transaminase (SGPT) (U/L)	Upto 45	418	75
Serum Glutamic Oxaloacetic Acid Transaminase (SGOT) (U/L)	0-40	440	60
Alanine phosphatase (U/L)	Upto 240	146	180
Gamma-Glutamyl transferase (U/L)	8-61	128	130
Total protein (g/dL)	6.4-8.3	4.6	5.02
Serum albumin (g/dL)	3.5-5	1.7 g/dL	2.85

Urea (mg/dL)	13-43	22.4	24
Creatinine (mg/dL)	0.7-1.2	0.77	0.45
Sodium (meq/L)	136-145	137	135
Potassium (meq/L)	3.5-5.3	4.2	4.7
Procalcitonin (ng/mL)	<0.5	0.14	0.09
Dengue ns1/IgM/malaria parasite scrub typhus/Typhidot		Negative	
Blood culture		Sterile	
Erythrocyte sedimentation rate-(mm in 1st hour)	<20	35	
High sensitivity C-Reactive Protein (Hs-CRP)	<6	22	
Urine routine Pus cell/Epithelial Cell/Red Blood Cells/Cast/Crystal		NIL	
Urine culture		Sterile	
24 hour urine protein	30-300 mg/24 hr	300 mg/24 hour	
Electrocardiography (ECG)		Non specific ST-T changes with sinus tachycardia	Normal sinus rhythm
Troponin I		negative	
Creatine phosphokinase (CPK-MB) (Unit/lit)	26-192	217 Unit/lit on first day, 446 Unit/lit on second day,1974 Unit/lit on third day	
Brain natriuretic peptide level (pg/mL)	<125	>6000	
Creatine Phosphokinase-N Acetyl Cysteine (CPK-NAC) (IU/L)	20-215	2520 IU/L on first day, 6620 IU/L on third day	208 IU/L
Lactate Dehydrogenase (LDH) (unit/L)	240-480	1547	
Thyroid Stimulating Hormone (TSH) (mIU/L)	0.5-5	2.94	
FT4 (ng/dL)	0.7-1.9	0.9	
Cerebrospinal Fluid (CSF) study	0-5 cells Acellular protein: 15-40 g/dL sugar: 45-80 mg/dL	Acellular protein 33g/dL, sugar: 43 mg/dL	
Electromyography (EMG)		Increased spontaneous activity, polyphasic motor unit action potential suggestive of myopathy	
Muscle biopsy			*Trichinella* larva

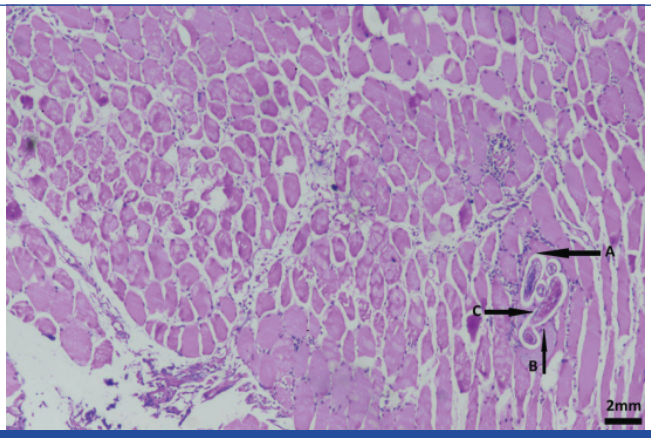
[Table/Fig-1]: Baseline investigations and after treatment.

The above investigations on day 1 of her visit, along with history, examination were suggestive of inflammation along with eosinophilia, and myositis with myocarditis. Cerebrospinal Fluid (CSF) did not reveal any albumin-cytologic dissociation, hence authors ruled out Guillain Barré syndrome. Muscle biopsy was planned to rule out immune-mediated myopathy as well as infectious myositis. During hospitalisation on day 3, she developed cardiogenic shock, her troponin I was negative, but Creatine Phosphokinase-Myocardial Band (CPK-MB) was highly raised, which led to the diagnosis of severe myocarditis leading to cardiogenic shock and was managed aggressively with intravenous (i.v.) fluid and ionotropic support. Two Dimensional (2D) Echocardiogram revealed global hypokinesia and ejection fraction of 20% [Table/Fig-2], however, it was advised to be repeated once tachycardia resolved. Given the severe myositis and cardiac involvement, she was started on i.v. methylprednisolone 1000 mg in 100 mL normal saline over two hours for three days (from third day of admission), and the patient responded dramatically. After methylprednisolone therapy, shortness of breath, with single breath counting improved. Her muscle enzymes (Creatine Phosphokinase-N Acetyl Cysteine (CPK-NAC) -208 (IU/L) and transaminases (Serum Glutamic-Pyruvic Transaminase (SGPT) -75 U/L, Serum Glutamic Oxaloacetic Acid Transaminase-60 U/L) also came down [Table/Fig-1]. She could get up from lying down position and walk alone by eighth day. She was continued on oral steroids 40 mg/day (1 mg/kg).



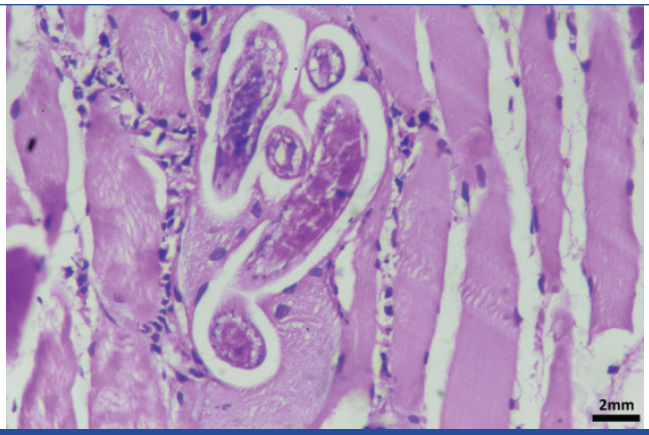
[Table/Fig-2]: Apical four chamber view of Echocardiography image during cardiogenic shock.

As the patient had severe weakness and severe tenderness in both lower limbs, a muscle biopsy was done from the lower thigh at quadriceps insertion. Previous literature also indicate biopsy to be done at insertion site of involved muscle [1]. Muscle biopsy was done on fifth day of hospital admission showed bundles of skeletal muscles with foci of lymphocytes and macrophages infiltrating the myofibers; one focus showed larvae enclosed within a membrane [Table/Fig-3,4]. Nucleated stichocytes, larva within the collagen capsule identified as *Trichinella* larva, keeping background clinical history of probable Trichinellosis, and epidemiology pattern. After a confirmed diagnosis of Trichinellosis by muscle biopsy, antihelminthic (albendazole) was also started at a dose of 400 mg twice daily orally from day 12 and was discharged on tab. albendazole 400 mg twice daily for a total 14 days duration and prednisolone (1 mg/kg) 40 mg daily and tapered off over next 14 days.

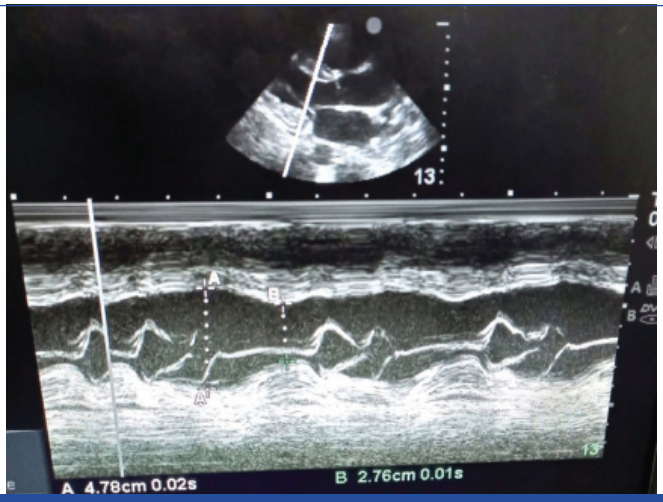


[Table/Fig-3]: A 10x magnification Histological section (Haematoxylin and Eosin (H&E) Staining) of muscle biopsy showing inflammatory infiltrates (A), collagen capsule of a cell (B), and a section of the muscle larva (C).

On follow-up after two months, she was doing well without any myalgia, and cardiac evaluation by 2D Echocardiography revealed normal ejection fraction of 55% [Table/Fig-5]. Her CPK-NAC was 70 IU/L, and transaminase was normal (SGOT-35 IU/L, SGPT-37 IU/L).



[Table/Fig-4]: A 40x magnification histological section of the muscle larva of *Trichinella* sp. stained with haematoxylin and eosin.



[Table/Fig-5]: Motion (M) mode image showing normal Left ventricular internal diameter end systole (LVIDs)=2.76 cm, and Left ventricular internal diameter end diastole (LVIDd)=4.78 cm on follow-up.

DISCUSSION

Trichinellosis is a zoonotic disease caused most commonly by *Trichinella spiralis* [1]. *Trichinella* infection in humans are often reported by eating raw or undercooked meat of infected domestic pigs, wild boars, wild pigs, horses, bears, walrus seals, and foxes [2,3]. Most of the infections are subclinical and remain undiagnosed [4]. In India, the infection of *Trichinella* is not uncommon, but diagnosis becomes difficult because of non specific symptoms of the disease. The number of reported cases has dropped significantly and has been around 20 cases every year from 2008 to 2010 [5]. Very few patients manifest a severe form of gastroenteritis, myositis, myocarditis and even may cause death [1,3,6,7]. Even tasting minimal amounts of undercooked meat during preparation or cooking may cause a risk of infection [8].

In India, the source of infection remains undercooked pork in most cases reported till now [4,9-14]. The life cycle of *Trichinella* starts with the consumption of undercooked pork by a human being that contains muscle larva followed by liberation of the encysted larva by digestive acids and pepsin. Later these larvae invade the small intestine mucosa, and there it matures from larvae to adult worms. The enteral phase of *Trichinella* is usually asymptomatic, lasts from 1 to 8 weeks, but abdomen pain, diarrhoea, nausea, and vomiting may occur. After maturation from larvae to adult, the female worms release newborn larvae, and around one week after this, they migrate to the striated muscle via the circulation, and they lodge in the striated muscles where they develop into muscle larvae encapsulated in the nurse cell. Depending upon the infectious dose and species of parasite, the incubation period of *Trichinella* ranges from one to two days (gastrointestinal phase) to two-eight weeks (parenteral phase) or may be more. Migrating *Trichinella* larvae lead to marked local and systemic hypersensitivity reactions together with eosinophilia [4,15].

The diagnosis is often by persistent peripheral eosinophilia associated with typical presentation fever, myalgia, periorbital swelling, oedema, and chest pain; almost always associated with elevated muscle enzymes, and cardiac biomarkers. Most commonly used test is by serologic testing by Enzyme Linked Immuno-Sorbent Assay (ELISA) or Indirect Fluorescent Antibody Test (IFAT) [4]. The detection of specific anti *Trichinella* antibodies, are of greater diagnostic value. The Immunoglobulin G (IgG) can generally be demonstrated 12 to 60 days. The mainstay of treatment is antihelminthic agents, however severe cases like myocarditis, myositis require aggressive glucocorticoid treatment. The differential diagnosis should include immune mediated myopathy, pyomyositis [10]. Complications like acute cardiac failure, cardiogenic shock, myositis, respiratory failure can develop. *Trichinella spiralis*-associated myocarditis does not result due to the direct larval invasion of the myocardium with encystation, but is caused most likely by an eosinophil-enriched inflammatory response which leads to eosinophilic myocarditis. Therefore, early and aggressive management is the key to successful treatment [1,3,16].

The present case represents a very severe fulminant manifestation of trichinellosis, as the symptom onset to disease progression happened within seven days and patient developed life-threatening cardiogenic shock, which makes the case different from previously reported cases where the presentation is indolent over three months duration. Both cases were managed with steroids and antihelminthics [3,17]. Also, the temporal association of antecedent acute gastroenteritis followed by muscle weakness and eosinophilia in peripheral smear strongly suspect infectious myositis as a cause [18]. A similar outbreak happened in April 2011 after uncooked pork consumption in a common feast [4]. Several cases have been reported causing severe myocarditis, pancarditis, myocardial infarction [6,19], encephalitis [6], myositis [20], and rarely death.

CONCLUSION(S)

Infectious myositis must always be kept in differential diagnosis of patients presenting with acute onset myositis with myocarditis and should be treated aggressively with steroid and anthelminthic.

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