

# A 12-year-old Child with Trichinellosis, Pyomyositis and Secondary Osteomyelitis

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## ABSTRACT

Trichinellosis is a parasitic infestation affecting the skeletal muscles. Cases of trichinellosis in humans have been reported from most regions of the world. However, a review of literature revealed only two reported cases of human trichinellosis in India. Further, a diagnosis of superimposed pyomyositis in trichinellosis with secondary osteomyelitis has not been reported to our knowledge. This article reports this rare case presentation in a 12-year-old child. Timely intervention helped prevent long-term morbidity in our patient. In our case report, we also discuss in detail the pathogenesis of such a condition and discuss the role of imaging modalities and an early magnetic resonance imaging (MRI) to diagnose the condition and start an early treatment.

**Key words:** Osteomyelitis, Pyomyositis, *Trichinella spiralis*, Trichinellosis

## INTRODUCTION

Trichinellosis is caused by tissue-dwelling helminth of the species *Trichinella*. It is acquired by the consumption of raw or uncooked pork products containing encysted larva. The larvae affect striated skeletal muscle cells. The presenting complaints most commonly include myalgia, fever, rash and fatigue. Arthralgias and cardioneurological symptoms occur rarely, but the parasite has not been known to directly infect the bone. Pyomyositis is primary infection of the skeletal muscle, usually caused by *Staphylococcus aureus*, and is endemic in tropical areas. To our knowledge, there have been only two reported cases of human trichinellosis in India.<sup>[1,2]</sup> This article presents a rare case of human trichinellosis in India, which predisposed to pyomyositis and secondary osteomyelitis. We also discuss in detail the pathogenesis of such a condition and discuss the role of imaging modalities and an early magnetic resonance imaging (MRI) to diagnose the condition and start an early treatment.

## CASE REPORT

A 12-year-old female child from Indian Gangetic belt presented with complaints of fever and pain in right thigh since 3 weeks. Fever which responded to antipyretics was of moderate degree with documentation not more than 101°F. The febrile episode was intermittent with no diurnal variation or associated chills and rigors. Two days after the onset of fever, the patient had reported pain and swelling in right distal thigh, with restricted movement of right knee due to pain. Patient had complaints of generalized myalgia in the initial week of fever. On further direct questioning, the patient gave a history of patchy red rash over the front and medial aspects of thigh on the 2<sup>nd</sup> day that lasted for around 6–8 hours. There was neither a history of trauma or photosensitivity nor any symptomatology localizing to respiratory, urinary or central nervous system. Further probing confirmed absence of any antibiotic treatment for fever till the time of admission. The patient hailed from a low socioeconomic background, was a non-vegetarian and occasionally used to consume pork. The patient had consumed hamburger in the past 1 month, following which she had an episode of diarrhea, dyspepsia and myalgia for 4 days. But the episode was self-limiting and the patient improved on her own without taking any medications. However, none of the relatives of the patient or any other person in the region had reported similar complaints.

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On examination, tenderness in the right thigh reaching up to the knee joint with a mild local rise in temperature was found. Knee movements were painful and restricted. Palpation of inguinal lymph nodes revealed lymphadenopathy. However, systemic examination including examination of other joints was found to be within normal limits.

X-rays of the right femur revealed grossly normal bony architecture but altered soft tissue shadows in the surrounding thigh musculature [Figure 1]. Laboratory investigations revealed an elevated WBC count ( $13,700/\text{mm}^3$ ) with eosinophilia of 10%. However, blood cultures were found to be sterile. A finding of eosinophilia led us to investigate for the serological markers of various parasitic infections. The serology tests were found to be positive for IgM antibodies to *Trichinella* species. *Trichinella spiralis* was suspected since it is the predominant *Trichinella* species in the region.<sup>[1,2]</sup> An ultrasound done at the same time revealed bulky and heterogeneous deep muscles of the posterolateral aspect of mid and lower thigh with a multiloculated fluid collection suggestive of myositis. This was followed by an MRI [Figure 2] that was done 2 weeks later which highlighted signs of acute osteomyelitis of lower end of femur with large collection of abscess in deep posterolateral aspect of thigh along with infiltration of adjacent muscles.

A serous discharge was obtained on needle aspiration, which on microscopy showed occasional pus cells; however, gram stain, acid-fast bacilli (AFB) and culture sensitivity were found to be negative. Nevertheless, a provisional diagnosis of acute osteomyelitis was kept and the patient was taken up for debridement and corticotomy. Intraoperatively, the muscles appeared to be edematous, bulky and unhealthy with minimal seropurulent discharge. Serous discharge amidst marrow fat was expressed on corticotomy. The entire area was drained and tissue specimens were harvested intraoperatively for histological and microbial evaluation. A detailed microbial work up revealed findings summarised in Table 1:

- Histopathologic examination of muscle biopsy revealed parasitic infection with eosinophilic infiltrates, and a possibility of parasitic cyst was kept on the basis of histological findings [Figures 3a-c]. Considering the isolation of *Trichinella* larvae as reported by the pepsin digest muscle biopsy study and a histological and laboratory picture suggestive of parasitic infestation, a diagnosis of *Trichinella* predisposing to bacterial pyomyositis was made.
- Considering the high prevalence of staphylococcal pyomyositis and osteomyelitis, the patient received 4 weeks of empirical intravenous cloxacillin along with ceftriaxone following surgical drainage and debridement. This was followed by additional 2 weeks



**Figure 1:** X-rays of the patient at presentation, showing grossly normal bony architecture



**Figure 2:** MRI of lower end of femur suggestive of acute osteomyelitis with large collection of abscess in deep posterolateral aspect of thigh along with infiltration of adjacent muscles

**Table 1: Results of Microbiological Work-up**

| Pus cells                                    | Positive (occasional) |
|--|-----------------------|
| AFB  | Negative              |
| Gram stain                                   | Negative              |
| Culture sensitivity                          | Sterile               |
| Fungal smear                                 | Negative              |
| Muscle biopsy of pepsin digested preparation | Trichinella larvae    |

AFB: Acid-fast bacilli

of oral cephalosporins. Oral mebendazole in a divided total dosage of 600 mg per day was given for a period of 14 days to cover for the *Trichinella* infection.

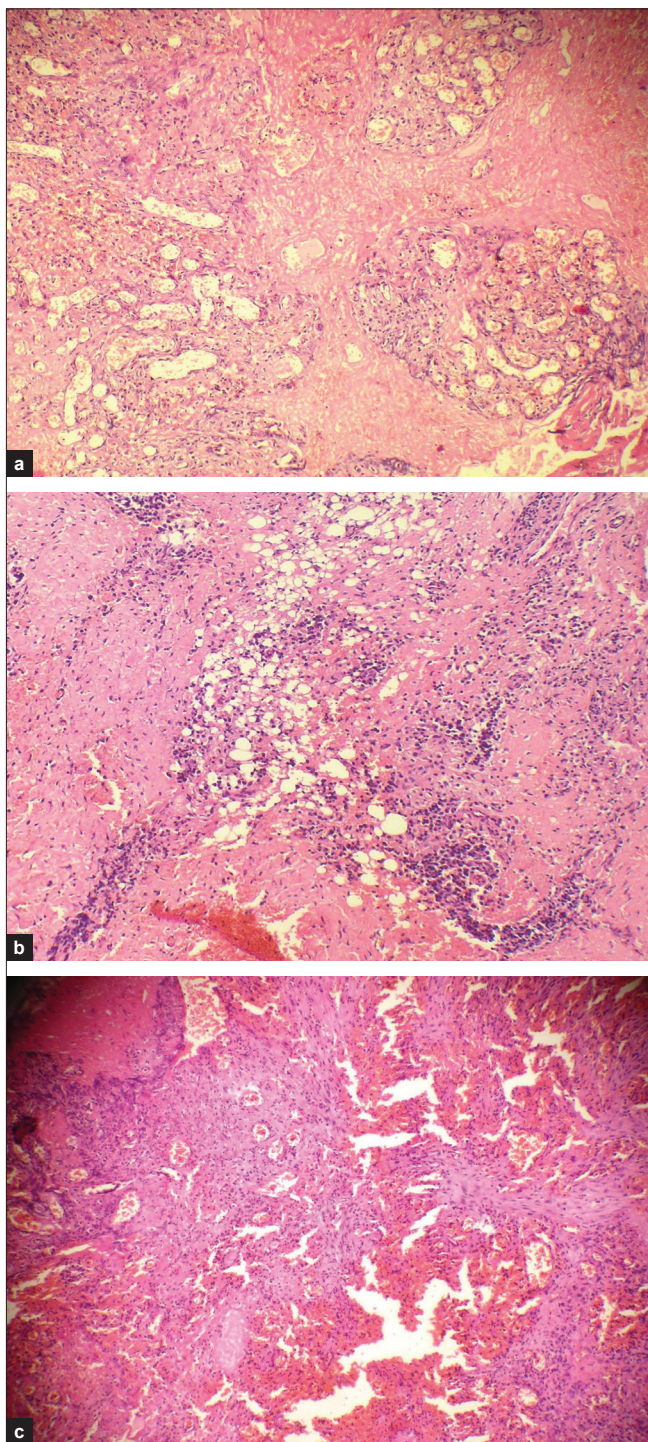
- The patient's general condition improved and fever subsided gradually over 1 week. Early range of motion at knee and mobilization with full weight bearing was initiated immediately after the surgery. Follow-up after 6 months showed the patient to be disease free with no complaints in playing or carrying out her day-to-day activities.

## DISCUSSION

After an extensive workup, our case was narrowed to a differential diagnosis of either acute hematogenous osteomyelitis with secondary pyomyositis or an unusual manifestation of *Trichinella* predisposing to pyomyositis and secondary osteomyelitis. The prevalence of staphylococcal osteomyelitis in children in tropical countries points toward bacterial etiology. However, in our case, the child never received any antibiotics prior to admission and neither were we able to isolate any bacteria in culture studies. The serous nature of the pus drained in this case was also not typical of bacterial pathology which presents with frank pus. Studies have reported that in almost 30% of cases of osteomyelitis with secondary abscess, no organism is isolated on cultures, and the likely reason for this is believed to be the low standards of laboratory facilities in tropical areas. However, our patient presented to a tertiary center institute with state-of-the-art laboratory facilities and isolation techniques. This led us to postulate *Trichinella* as one of the possible etiologies in the case. Although a Western blot analysis performed at this stage could have been confirmatory, this was not done by us and our diagnosis primarily rested on the isolation of *Trichinella* larvae in the pepsin digested muscle biopsy study.

We believe that this is one of the very rare instances when such an atypical presentation of trichinellosis causing pyomyositis with secondary osteomyelitis has been reported. Bennet *et al.*<sup>[3]</sup> in 1928 reported a case of sclerosing osteomyelitis caused by trichinellosis, while Steel *et al.*<sup>[4]</sup> in 1964 reported osseous complaints and myoperiosteal larval infiltration by *Trichinella*.

Trichinellosis, also called trichinosis or trichiniasis (Trich from Greek thrix meaning hair), is an infestation caused by nematodes of the genus *Trichinella*, most commonly *T. spiralis*. Infection is initiated by ingestion of viable larvae in raw or undercooked meat. Digestive action liberates the larvae. Newborn larvae penetrate the intestinal wall, enter the lymphatic system, and move via the bloodstream to areas of implantation. The larvae travel by capillaries to various organs such as the retina, myocardium, or lymph nodes; however, only larvae that migrate to skeletal muscle cells survive and are encysted. Once in the cell, they alter the cellular activity to turn the individual cells into “nurse cells” in which the larvae encapsulate. The development of a capillary network around the nurse cell completes the encystation of the larvae. The symptoms in the enteric phase include those of dyspepsia, diarrhea, nausea, and heartburn. In the parenteral phase, the body’s inflammatory response results in edema, muscle pain, fever, and weakness.



**Figure 3:** Histological profile. (a) The microphotograph showing a fibrocollagenous tissue with extensive small blood vessel and capillary dilatation as well as congestion. The dilated capillaries are heavily surrounded by dense polymorphonuclear cells and debris. The overall picture is of a granulation tissue. Very occasional eosinophils are also seen. (b) One of the portions of the same tissue showing predominantly a lymphomononuclear inflammation admixed with some polymorphs and debris. The inflammatory foci are also infiltrated by adipose cells. (c) Other focuses showing a mixed inflammatory infiltrate with few congested capillaries. The inflammatory cells are composed of neutrophils and lymphomononuclear cells in a background of hemorrhage

Cardioneurologic symptoms consisting of encephalopathy, focal deficits and myocarditis represent the severe life-threatening forms which occur very rarely. In the heart, acute inflammatory changes are found during the early stages in the form of a patchy but scattered interstitial myocarditis; however, the larvae do not encyst within the myocytes. Invasion of CNS is reflected by a diffuse lymphocytic and monocytic infiltration in leptomeninges and development of focal gliosis.

Diagnosis of trichinellosis in a person assumes high significance considering the potential of the parasite to cause multiple outbreaks and epidemics. It is very important to be sure that the patient is the single human case reported. Also, it is equally important to trace the origin of infected meat and possible worm burden. To our knowledge, our patient was the only case of trichinellosis reported in the area during that time interval. Considering that the diagnosis of trichinellosis was delayed, it was not possible for us to trace the source of infected meat.

Pyomyositis denotes an abscess of skeletal muscle that occurs either spontaneously, as in primary pyomyositis, or secondary to a penetrating injury or local spread from an adjacent soft tissue or bone. Primary myositis is also termed tropical pyomyositis, myositis tropicans, tropical skeletal muscle abscess, and tropical myositis. Although the classical presentation is with muscle abscess, the hallmark of the disease is not an abscess but a finding of myositis on the biopsy specimen of involved muscle.<sup>[5]</sup> Trauma,<sup>[6,7]</sup> immunological disorders and parasitic infestation<sup>[8]</sup> have all been postulated to favor primary pyomyositis. *S. aureus* is the primary pathogen in 90% cases. Group A streptococci are probably the next in frequency. Less common causes include groups B, C, and G streptococci, pneumococci, *Haemophilus influenzae*, *Aeromonas hydrophila*, *Fusobacterium* sp., *Bartonella* sp., Gram-negative enteric flora, and anaerobes. Although rare, tuberculous and nontuberculous mycobacterial pyomyositis has also been described in individuals who are immunocompromised.<sup>[9]</sup> There are also occasional reports of *Salmonella*<sup>[10]</sup> and gonococci<sup>[11]</sup> as causative organisms, usually secondary to disseminated infections with these bacteria. Parasitic (filaria, nematodes) and viral causes (HIV, Herpes, Picorna virus, Arbo virus) have also been reported.<sup>[12]</sup> A recent study has demonstrated a possible link between pyomyositis and toxocariasis.<sup>[13]</sup>

Human trichinellosis has been rarely reported from India. A review of literature revealed only two such case reports from India.<sup>[1,2]</sup> In fact, we believe that most of the physicians in India are practically unaware of the condition

due to its rarity. As such, a diagnosis of trichinellosis is likely to be missed in countries like India. The aim of our report is to spread awareness amongst the Indian physicians regarding the existence of this entity.

In our patient, trichinellosis with pyomyositis followed by secondary involvement of bone seems to be a logical explanation for the order of events. The child presented to us late and hence we do not have initial imaging to suggest any such evolution. But considering that the primary predilection for *Trichinella* is muscle tissue and that presentation was delayed by almost 3 weeks, osteomyelitis seems secondary to pyomyositis. There are varying reports in the literature where osteomyelitis had evolved as a complication of pyomyositis.<sup>[14,15]</sup> The underlying causes for such an association might include a delay in treatment, immunosuppression and atypical organisms causing the disease. In our patient, X-rays obtained around 3 weeks after the onset of symptoms failed to show any features of osteomyelitis, although soft tissue changes suggestive of pyomyositis were present. However, after another 10 days, the MRI showed acute osteomyelitis with pus collection in muscles. This further points toward secondary involvement of bone in our patient as initially the X-rays were normal. If pus was secondary to osteomyelitis, the X-rays would have shown signs of osteomyelitis.

Osteomyelitis as a differential diagnosis for pyomyositis is often described in literature, but there are very few reports of pyomyositis leading to secondary osteomyelitis. In one study by Block *et al.*,<sup>[16]</sup> 8 of the 11 patients with staphylococcal pyomyositis developed osteomyelitis as a complication. In another study by Chiu *et al.*,<sup>[17]</sup> 5 out of 24 patients with bacterial pyomyositis were seen to develop secondary osteomyelitis. Our case is unique for two reasons. First, osteomyelitis developed as a complication of pyomyositis, and second, trichinellosis appeared to be a predisposing factor to primary pyomyositis. It is well documented that parasitic infestations with filaria and *Toxocara* predispose to pyomyositis,<sup>[18,19]</sup> but *Trichinella* infection predisposing to primary pyomyositis is being reported for the first time. The diagnosis of trichinellosis is supported by the positive serology for IgM and the larval findings in pepsin digested muscle specimen. Since we did not do the Western blot analysis, we believe that the diagnosis was only highly suggestive and not fully proven. However, there is no microbiological evidence for the etiological agent for pyomyositis in our case as bacteriological workup was inconclusive. *Trichinella* was the only pathogen isolated. Although primary *Trichinella* pyomyositis could be considered as a possible diagnosis, such a condition seems highly unlikely considering the fact

that pus formation is unusual in the course of trichinellosis. Moreover, we do not have an evidence to demonstrate *Trichinella* in bone specimen as causative for osteomyelitis. In the literature, barring a few isolated cases, secondary bone involvement by *Trichinella*<sup>[1,2]</sup> has not been reported in detail.

Usually, X-ray features of osteomyelitis appear by the 2<sup>nd</sup> week of onset of complaints. MRI in this case was done almost 4 weeks after the onset of symptoms. We believe that MRI is an important modality that helps in picking up early involvement of bone secondary to pyomyositis. In fact, serial MRI would be an ideal way to see for any evolution of pyomyositis into osteomyelitis. From our experience, we advocate the use of MRI in cases where an abscess is located adjacent to bone. Isotope bone scans may be performed if MRI is not available. However, MRI is more sensitive and is also a superior imaging modality for showing the anatomical details. If concomitant osteomyelitis is found, treatment has to be tailored accordingly.

### CONCLUSION

Human trichinellosis has been reported very rarely from India and many physicians in the region are practically unaware of this entity. Further, *Trichinella* predisposing to pyomyositis and then evolving into secondary osteomyelitis is a rare atypical presentation not described much in the literature. Negligence and late presentation in our patient led to the development of osteomyelitis secondary to pyomyositis. We believe that an initial MRI is the diagnostic modality of choice to make an accurate diagnosis at an early stage. An early diagnosis will in turn avoid progression of the disease and reduce the associated morbidity due to further complications.

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