

Case Report

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Mysterious Cause of Prolonged Low Back Pain: Isolated Iliococcygeal Tubercular Myositis- A Rare but Existing Clinical Entity

Richmond R Gomes*

Department of Medicine, Ad-din Women's Medical College Hospital, Dhaka, Bangladesh

***Corresponding author:** Richmond Ronald Gomes, Associate Professor, Department of Medicine, Ad-din Women's Medical College Hospital, Dhaka, Bangladesh.

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Abstract

Tuberculosis remains one of the deadliest diseases in the world. The World Health Organization (WHO) estimates that each year more than eight million new cases of tuberculosis occur and approximately three million people die from the disease. Isolated tuberculous myositis (ITBM) is a rare entity. literature. This diagnosis is very frequently missed due to its rarity and atypical clinical presentation, which leads to delay in treatment. A high suspicion index and a prompt intervention is a must for such a rare clinical presentation of extra pulmonary tuberculosis. This entity should be considered as differential diagnosis for prolonged low back pain with fever. We report the case of isolated tuberculosis of the left sided iliococcygeal muscle without any evident primary focus in a 52-year old immunocompetent female.

Keywords: Tuberculous myositis; Extra pulmonary tuberculosis; Iliococcygeal muscle

Introduction

Tuberculosis (TB) has remained a major global and public health burden and a leading cause of deaths worldwide [1]. The global TB incidence in 2022 increased by 11%, compared to the incidence in 2008. Although Mycobacterium tuberculosis infection mainly manifests as pulmonary TB (PTB), TB can affect any organ or tissue. Extrapulmonary TB (EPTB) accounts for approximately 15% of new TB cases, and the EPTB incidence increased by 33% from 2013 to 2022. Musculoskeletal EPTB is uncommon (1%~3% of all TB cases), and spondylitis, arthritis, and osteomyelitis are responsible for most EPTB cases [2]. TB myositis has been sporadically reported, and can occur through contiguous infection or he-

matogenous spread; muscle involvement is generally secondary to disseminated TB or adjacent skeletal EPTB. However, TB of only the skeletal muscle, which is known as isolated TB myositis (ITBM), is very rare. Therefore, since 1886 when the involvement of lower leg muscle was first described, there has been little information on the clinical features of ITBM and its associated conditions.

Case Report

Mrs. X, 52 years old muslim teacher, not known to have diabetes, hypertension, bronchial asthma or coronary artery disease presented to us with the complaints of fever for 2 months and low back pain for the same duration. Fever was low grade, intermittent with

mostly evening rise (highest recorded temp was 101oF). Fever subsided with anti-pyretic with profuse sweating. Fever was not associated with headache, vomiting, altered consciousness, cough, chest pain, hemoptysis, dysuria. Fever was associated with non-radiating low back pain which was initially mild but worsened over time. There was no definite inactivity stiffness. There was also no significant history of preceding diarrhoeal illness or urethral discharge. She denied any other joint pain, sole pain, skin rash or patch, red eye, oral or genital ulcer. Family history of such type of illness or contact with patient with active tuberculosis were not found. There was no significant personal, past or recent travel history. On query she had anorexia and had unintentional weight loss of 5 kgs in last two months. On examination, patient was ill looking, cooperative, hemodynamically stable. Temperature was raised (100.8 o F). Other general examination findings were non-significant. Locomotor system examination revealed negative SLR. Patrick test (for both hip joints) sacroiliac compression or extraction test revealed no

abnormalities. There was localized lower spinal tenderness. Initial lab work up showed normocytic normochromic anemia (Hb% 10.1 gm%, MCV89.9 Fl, MCH 28.8 pg), neutrophilic leukocytosis (Total count 20×10^3 , N- 78.8%). ESR and CRP was raised to 95 mm in 1st hour and 320 mg/L (normal upto 5 mg/L) respectively. Urine routine examination, SGPT, serum creatinine, serum electrolyte, serum uric acid level was normal. X ray of lumbosacral spine showed mild degenerative changes. Blood culture revealed no growth and HLA B-27 report came negative. So provisional diagnosis of spondyloarthritis was made and she was started treatment with systemic corticosteroid and muscle relaxant therapy. On 4th day of starting treatment, her condition worsened and she became bed bound. MRI of lumbosacral spine with both hip joints and both SI joint was advised which revealed mild thecal sac indentation at L4/5 and L5/S1 level and normal hip joints without any features of sacroiliitis (Figure 1 and Figure 2).

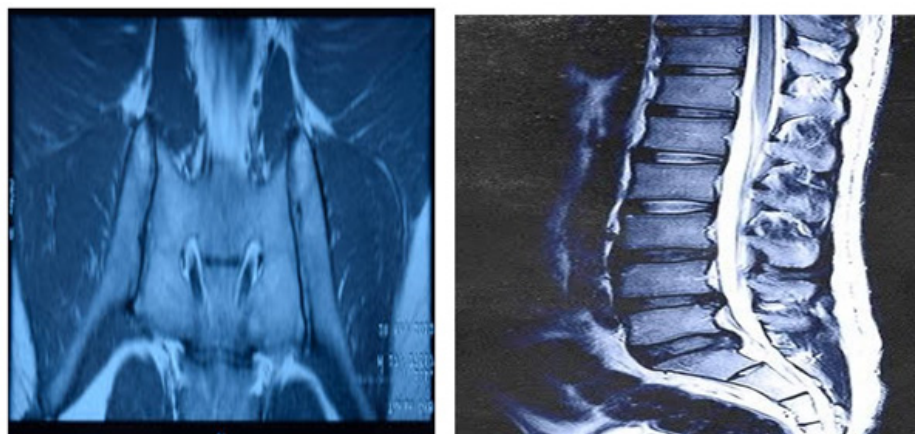


Figure 1 and Figure 2: Normal both SI joints and mild thecal sac indentation at L4/5 and L5/S1 level.

As the patient remain febrile & there was no evidence of spondyloarthritis, corticosteroid therapy was stopped. She was empirically started with meropenem and linezolid, assuming any underlying occult infective pathology. Despite treatment with antibiotic for 5 days, there was no significant clinical improvement as evidenced by persistent fever & no improvement of CRP (320mg/dl to 280 mg/dl). After proper counselling with patient's attendant, MRI of Pelvis was planned which revealed Diffuse T2 hyper intensity over left Ilio-coccygeus muscle. Features suggestive of infective myositis (Figure 3).

Serum CPK was mildly raised 200u/L (normal35-170u/L). Mantoux test became positive)14 mm after 72 hours). Both surgery & Orthopedics consultations were sought. There was a plan to do muscle biopsy for histopathology and Gene X pert TB, but patient party was refused to do so. Considering prolong febrile illness & non-responded to broad spectrum antibiotic therapy, empirical

anti tubercular treatment was started according to body weight with oral prednisolone (1 mg/kg/day). After 10 days of starting anti tubercular treatment, Fever was subsided & mild improvement of low back pain. So, we planned to discharge her with anti-tubercular medications (rifampicin, isoniazid, ethambutol and pyrazinamide in fixed dose combination) and tapering oral prednisolone. She was advised for out patient door follow up after 1 month. During her follow up visit after 1month, there was no fever & significant improvement of low back pain. There was also anti- tubercular drug induced adverse events. So, we planned to continue anti tubercular drug for 9 months (intrinsic phase with four drugs for 2 months followed by continuation phase with rifampicin and isoniazid for next seven months) and oral steroid for 8 weeks. She was also advised to do repeat MRI of pelvis at the end of her treatment. Repeat MRI of pelvis at 9 month revealed normal left ilio-coccygeal muscle (figure 4). Patient was afebrile, no back pain and she also gained 8 kg weight in last 9 months (Figure 4).

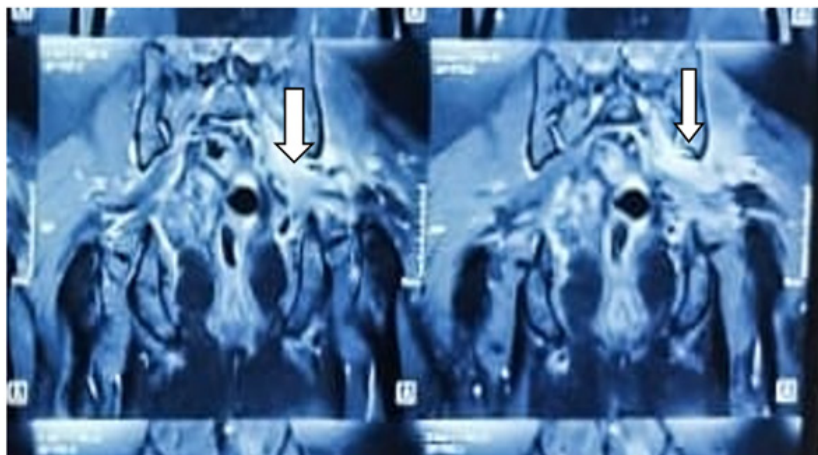


Figure 3: Hyper intensity over left Ilio-coccygeus muscle suggestive of infective myositis (white arrow).

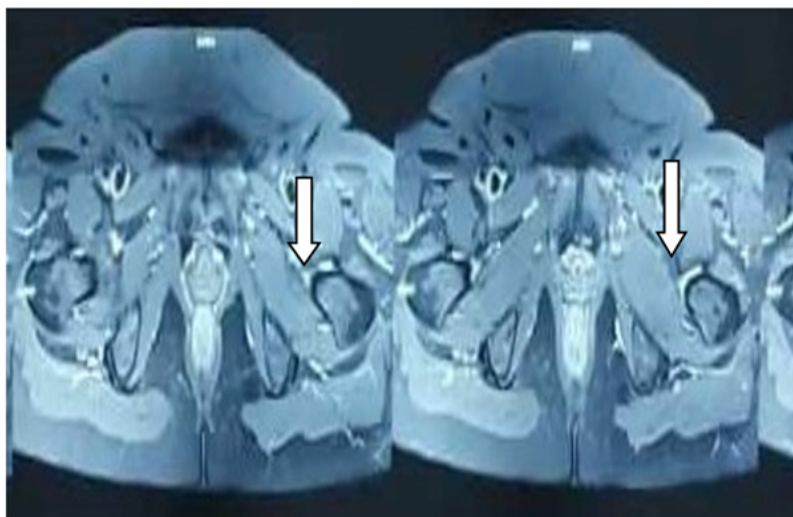


Figure 4: MRI of pelvis showing normal left ilio-coccygeal muscle(white arrow).

Discussion

Pyomyositis is a purulent infection of muscle that is generally the result of hematogenous spread [3]. Typically, the infection is known to occur in the tropics in otherwise healthy individuals with no comorbidities. Those afflicted in more temperate regions tend to have severe underlying comorbidities or are immunocompromised in some way [4]. The most common organisms implicated in pyomyositis include *Staphylococcus aureus* as well as, increasingly, MRSA. Group A streptococci are also common with gram-negative bacilli and pneumococci, with non-group A streptococci occurring less often [4]. There have also been instances of mycobacterial-induced pyomyositis [5]. It rarely occurs without bony involvement.

Heffner in 1977 first described focal myositis as a local, self-lim-

iting inflammatory lesion in skeletal muscle having unknown etiology [6]. As skeletal muscle is not a favorable site for growth and survival of mycobacterium tuberculosis, tuberculosis in soft tissue without an underlying pathology is very rare. This is due to high lactate content, poor oxygen, rich vascular supply, highly differentiated state of muscle tissue and absence of lymphatic system [7]. Tuberculosis can spread directly to the skeletal muscles from bone through synovium of tendon sheaths, joints or by direct inoculation or rarely hematogenously. The diagnosis is often missed due to lack of typical signs and symptoms and rare incidence. The delay in diagnosis and treatment leads to wider spread, deformity and atrophy of the involved muscles. In our cases, tuberculous involvement in iliococcygeal muscle seems primary because there were no tuberculous foci in any other part of the body and no notion of

iatrogenic inoculation was noted. The involvement of the muscles coexists with pulmonary involvement in 30% of cases [8]. Pulmonary tuberculosis can be associated with rare locations such as tenosynovial or bursitis tuberculosis, but in both cases, mycobacterium tuberculosis was unusually isolated [9,10].

Clinical symptomatology associates pain and swelling of the affected muscle. Other constitutional symptoms of tuberculosis may or may not be present. The diagnosis of skeletal muscle tuberculosis remains a challenge for clinicians and requires a very high index of suspicion. The combination of clinical tests with radiological and cytopathological imagery, and a positive tuberculin skin test, provide strong clues pointing to a diagnosis of skeletal muscle tuberculosis. High degree of suspicion is needed for early diagnosis to avoid complications especially in endemic areas [5,11].

Although culture and histopathological examination is the gold standard for diagnosis, GeneXpert (Semi Nested Real Time PCR) can be used as an effective tool for rapid diagnosis. A negative ZN stain for AFB, normal chest radiograph, with no contact history of tuberculosis and no active tubercular foci does not rule out the diagnosis of tuberculosis. A high index of suspicion is required in a TB endemic country like Bangladesh, for early diagnosis and treatment. Imaging findings, including ultrasound, CT, and MRI, could identify lesions and sites for biopsy [12,13]. The evaluation should begin with ultrasonography. The tissue masses of the soft parts had a hypoechogenic appearance in most cases. Sonography allowed for differentiation between solid and liquid lesions, and some soft tissue tumors, hematomas, and cystic diseases could be distinguished [14]. However, ultrasound was a very sensitive but non-specific examination. In general, a soft tissue mass of unknown character in the lower extremity requires further workup via MRI, because this may confirm the size and nature of the mass more clearly than ultrasound, particularly in indicated tissue swelling and deeper tissue damage [15].

The differential diagnosis list for MT is broad, especially in the case of some patients presenting clinically with muscle nodules. The most common differentials include schwannomas, lipoma, liposarcoma, ganglionic cyst, focal myositis, and hydatid cyst of the muscle. Because of a suspicion of malignancy, exploration was undertaken and an open biopsy was conducted on the nodule in two patients. Focal myositis could also present with similar clinical and imaging presentation, but the pathological features such as inflammatory infiltration and muscle fiber atrophy, necrosis, and regeneration were not obvious in the muscle pathology of these two patients. In fact, during diagnosis, oncological vigilance should be maintained, because MT presentation is similar to that of some patients with tumors.

Treatment is based on antitubercular therapy (susceptible to organism) with a minimum of four drugs for a prolonged period. Isoniazid, rifampicin, pyrazinamide and ethambutol regimen is used. Surgical intervention can be an adjunct to antitubercular therapy [16]. The optimum duration of treatment is always debated; treatment should be prolonged to minimum nine months be-

cause the short antitubercular therapy may not be appropriate for extrapulmonary TB specifically for osseous involvements [16].

Conclusion

Muscle tuberculosis remains exceptional and its exact pathogenesis is not yet clearly explained. It should also be considered in the differential diagnosis of prolonged low back pain with fever especially in people born in tubercular endemic areas. Anti-tubercular therapy should be given as early as diagnosis is established to avoid atrophy and deformity of the involved muscle. The prognosis is good if well treated.

Conflict of Interest

None declared.

Acknowledgement

None.

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