Case report

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A rare case of primary tuberculous pyomyositis. Case report.

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Abstract

Tuberculosis involving the soft tissue as extending from adjacent bone or joint is well recognized. However, the primary tuberculous pyomyositis is rare. Due to atypical presentation the diagnosis is often delayed. We report one case of primary tuberculous pyomyositis of the thigh in an immunocompromised patient. Tuberculous myositis should be suspicioned, in immunocompromised patients with unexplained soft tissue swelling, especially in endemic area.

Keywords: tuberculosis, pyomyositis, corticosteroids, diabetes mellitus

Rezumat

Afectarea, prin contiguitate, a ţesuturilor moi în contextul unei infecţii tuberculoase osoase şi/sau articulare respectiv a unui abces rece de vecinătate, reprezintă o entitate clinică bine definită. Cu toate acestea (pio)miozita tuberculoasă primară este foarte rar întâlnită în practica clinică curentă. Manifestările clinice şi localizările atipice contribuie semnificativ la întârzierea diagnosticului. Prezentăm cazul unei paciente imunosupresate cu (pio)miozită tuberculoasă primară a coapsei drepte. Pentru stabilirea unui diagnostic de (pio)miozită tuberculoasă este nevoie de un înalt grad de suspinciune clinică în faţa oricărei tumefieri inexplicabile de ţesuturi moi, în mod particular în cazul pacienţilor imunosupresaţi din zonele endemice.

Cuvinte cheie: tuberculoză, piomiozită, corticoterapie, diabet zaharat

Introduction

Tuberculosis (TB) is considered as a "re-emerging disease", due to its resurgence and increasing incidence in the twenty-first century, particularly in immunocompromised patients [1]. About one-fifth of new diagnosed cases of TB have extrapulmonary lesion, of which about one-tenth involve the musculoskeletal system, mostly spondylitis, osteomyelitis, or arthritis [2]. Tuberculous myositis has been rarely described in the medical literature and its manifestations may mimic malignant or other inflammatory diseases [3,4]. TB can involve skeletal muscle by extension from the underling bone, neighbouring joints or tendon sheaths, or from cold abscess, by direct inoculation and by haematogenous dissemination [5]. Nevertheless TB of soft tissue without underlying bony pathology is very rare and the pathogenesis is still confusing [6].

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Case report

A 66 years old woman was admitted with a 2-week history of pain in the anterior and lateral aspect of the right thigh and right knee, fatigue, chills, fever, and altered knee range of motion Due to the pain the walking was impossible. She denied sweating, cough, haemoptysis, or loss of appetite. There was no history of recent TB contact and there were no known risk factors for HIV infection.

Past medical history included type 2 diabetes mellitus complicated with diabetic peripheral neuropathy, dyslipidemia, spondylarthrosis, and osteoporosis. One and half year prior to admission the patient was diagnosed with Polymyalgia Rheumatica (PMR) and corticosteroid

therapy (Prednisone 10 mg daily) and azathioprine (Imuran 100 mg daily), as steroid sparing agent, was started. At that time, rheumatoid arthritis, polymyositis, calcium pyrophosphate deposition disease, adult-onset Still's disease, hypothyroidism, monoclonal gammapathy, paraneoplastic syndrome and giant cell arteritis were ruled out.

On clinical examination she had fever (38.5 °C) but no rash or lymphadenopathy were noted. Musculoskeletal examination revealed an extremely tender and diffuse swelling of the anterolateral aspect of the right thigh. The overlying skin was warm and erythematous. The right knee was mildly swollen and tender. The examination of the right hip was normal. There was no spine tenderness or spinal movement restriction. On palpation of the abdomen, there were no physical abnormalities. Further examination of respiratory, cardiovascular and nervous system failed to delineate any abnormality.

Laboratory tests showed a non-specific inflammatory syndrome (erythrocyte sedimentation rate, ESR = 95 mm/1h, C-reactive protein = 4.8%), anemia (hemoglobin = 9.1 g/dL, hematocrit = 31%), neutrophilia (90%), hyperglycemia (183 mg/dL), hypercholesterolemia (231 mg/dL), hypertriglyceridemia (290 mg/dL) and elevation of muscle enzyme: creatine kinase, CK (609 IU/L), lactate dehydrogenase, LDH (489 IU/L) and aspartate aminotransferase, AST (64 IU/L). Repeated hemocultures (3 probes) were negative. Radiographs of chest and dorsolumbar spine revealed no abnormality and in particular no evidence of pulmonary or spinal TB. The X-ray examination of the right femur and right knee revealed no underlying bone lesions.

Ultrasound examination of the affected area (the lower half of right vastus lateralis muscle) depicted blurring/fading of normal muscular fibres appearance and associated muscle and (peri)fascial and subcutaneous edema (fig 1). No fluid collection was detected. No tendency towards fluid accumulation was observed in the next week of follow-up but edema become more expressed (fig 2). Ultrasound guided needle muscle biopsy was performed and the material was positive for acid-fast bacilli by Ziehl-Neelsen staining and four weeks later culture-positive for *Mycobacterium tuberculosis* in Lowenstein-Jensen medium. Histologic examination revealed muscle cells necrosis, fragmentation and loss of nuclei of muscle fibers, interstitial enlargement, "shadow" inflammatory cells (ghost-like remnants) and "nuclear dust" (karyorrhexis). Immunohistochemical analysis revealed LCA (leukocyte common antigen)-positive cells and a few CD68-positive cells (histiocytes). No lymphoma, sarcoma or neoplastic features were identified. Synovial fluid from the right suprapatellar bursa was negative for culture of bacterial pathogens, including *Mycobacterium tuberculosis*. No primary focus of TB was identified after thorough systemic and laboratory investigations. A final diagnosis of **primary tuberculous myositis** was established and antituberculosis therapy was started: isoniazid (INH), rifampin (RIF), pyrazinamide (PZA) and ethambutol (EMB).

Clinical evolution was unfavorable during the first 2 months, with persistent systemic symptoms, progres-

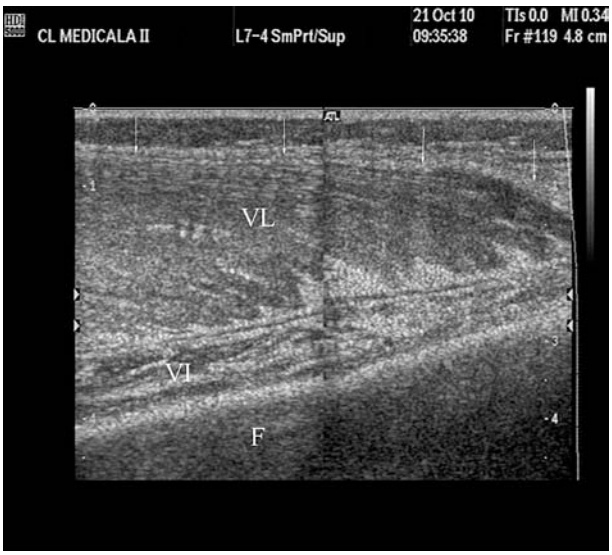


Fig 1. Longitudinal scan of the distal lateral tight. Note the blurring of the normal muscle structure of the vastus lateralis (VL) comparing with the structure of vastus intermedius (VI). VL is swollen and epimysium distended (arrows). F- femur.

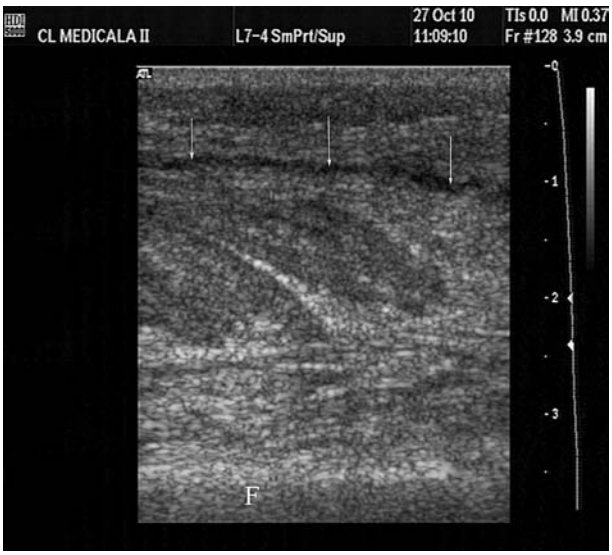


Fig 2. Longitudinal scan of the distal lateral tight, one week later as comparing with fig 1. Edema into the fascia appeared. No abscess is noted.



Fig 3. MRI examination, coronal T2 weighted image: edematous changes in the lower half of the vastus lateralis muscle, encapsulated fluid-filled homogeneous collection (C) with thin walls caudal fistulous tracts (arrow)

sive aggravation of the local signs and suspected drug-induced hepatitis. All potential hepatotoxic drugs were stopped (INH, RIF and PZA) and then restarted (in sequential fashion), once the AST level returned to normal. PZA was replaced by streptomycin (SM). MRI of the right thigh shows moderate edematous muscular enlargemet (T1W hypointensity and T2W hyperintensity) of the distal half of vastus lateralis, a fluid-filled homogeneous collection (T1W iso-intensity and T2W hyperintensity) surrounded by a very thin enhancing wall, with craniocaudal, laterolateral and anterosposterior diameter of 9.8 cm, 2.5 cm and 5 cm respectively, located in the lower third of vastus lateralis muscle. Multiple caudal fistula tracts in proximity of femoropatellar joint (fig 3). A diagnosis of **tuberculous abscess** was made and the patient was referred for surgical drainage. The post-surgical evolution was slowly favorable. The patient currently receives antituberculosis therapy and antidiabetic medication.

Discussions

Pyomyositis is the term used to describe a bacterial infection of skeletal muscle with abscess formation. The exact pathogenesis is still unknown, but factors like trauma, nutritional deficiencies, viral infections, septic load and parasitic infestations have been implicated as the predisposing factors [7]. Infection with *Mycobacterium tuberculosis* is an extremely rare cause for pyomyositis, particularly in immunocompentent patients. There are few case reports in the literature about TB pyomyositis found in HIV infected or renal failure patients, patients on chemotherapy or corticotherapy and chronic drug abusers [8,9]. The possible explanations for the rarity of muscle involvement in TB include the high lactic acid content of the muscle, absence of reticulo-endothelial and lymphatic tissue, highly differentiated state of muscular tissue, and its rich blood supply [10]. The infection by tubercula bacilli involves a single large muscle, most commonly the quadriceps femoris. Most of the authors have the opinion that the involvement of skeletal muscle is secondary to underlying bones, synovial sheats of nearby joints TB, either by direct inoculation or hematogenous dissemination [11]. Just reverse to this concept, some authors [12] have reported soft tissue (muscular) TB as the primary site for infection. In the absence of a direct spread from an adjacent primary focus, tuberculous pyomyosytis may be found in three circumstances: inoculation through needles and syringes contaminated with mycobacterium, idiopathically in an immunocompetent host and, as in our case, in immunocompromised patients. Involvement of the vastus lateralis muscle in our case seems primary, as there was no evidence of a tuberculosis focus elsewhere in the body.

Over the past decade pharmaceutical agents directed against TNF-α (infliximab, adalimumab and etanercept) have been widely and successfully employed for the management of rheumatoid arthritis, ankylosing spondylitis, psoriasis, psoriatic arthritis, juvenile idiopathic arthritis and inflammatoty bowel diseases. TB disease is a potential adverse reaction from treatment with TNF-α antagonists. The cardinal role of TNF-α in granuloma formation and clearance of intracellular microoraganisms is well known. The disruption of granuloma formation is thought to contribute to the increased risk of TB with these agents. TB infections associated with anti-TNF-α agents tend to reveal unusual clinical manifestations (multiple organs and atypical extrapulmonary pattern), including (pyo)myosytis [13,14].

In conclusions tuberculous myositis should be considered as one of the possible etiologies of muscular disease, especially in areas where tuberculosis is endemic. The basic step in the diagnosis is clinical suspicion (any atypical muscular swelling). With the increased reports of TB in immunocompromised hosts, typical clinical presentation is lacking. Diagnostic techniques like ultrasound and computed tomography/magnetic resonance imaging are very useful in diagnosis [15-17]. A muscle biopsy or aspiration with culture clinches the diagnosis.

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