

Cash Report

Forearm intramuscular tuberculosis of flexor digitorum profundus: a case report

Chandrakant C. Gharwade¹, Pranav G. Jawade², Shamol Banerjee¹, Ameya Bindu¹,
Al-Iqyan Fidvi J.¹, Shweta C. Gharwade³

Department of Pathology, Sri Ramachandra Medical College and hospital, Chennai, India

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***Correspondence:**

Dr. Chandrakant C. Gharwade,
E-mail: crgharwade@gmail.com

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ABSTRACT

Tuberculosis is one of the most commonly occurring infectious disorders in the developing countries like India, which literally can affect any organ and system. Primary tuberculous myositis is used to describe the clinical situation in which the most prominent lesion in the course of haematogenous tuberculosis involves the muscle and not the fact that the infection occurred primarily in the muscle. Tubercular pyomyositis is the least frequent extra spinal musculoskeletal tuberculosis reported in literature. We came across a 15 year old female without any known primary pulmonary or lymphoid foci who presented with a swelling and pain over right forearm since 3 months. After running all the radiological investigations and nerve conduction studies and taking under consideration the various differentials of the swelling, finally the swelling was excised and histopathology of the excised specimen was performed which suggested that the swelling was due to primary tuberculosis of the muscle and the underlying subcutaneous tissue and after that patient received anti tuberculosis regimen for 9 months and no recurrence of the disease was noted till date. This rare case is presented with review of literature.

Keywords: Intramuscular, Tuberculosis, MRI, Immunocompetant

INTRODUCTION

Tuberculosis is a worldwide occurring phenomenon, which has developed a global concern and has majorly affected most of the Asian and the African countries. Tubercular pyomyositis is the least frequent extra spinal musculoskeletal tuberculosis reported in literature.¹ The global incidence of Tuberculosis has drastically risen over the past decade and the World Health Organization (WHO) raised some serious concerns regarding this disease. Primary tuberculosis of skeletal muscle was first described in the year 1886 and is a rare, occurring phenomenon in 1% to 3% of all infected patients.² Musculoskeletal TB has a strong affinity for bones,

joints, muscles, tenosynovium and bursa and can mimic several other pathologies leading to a wide spectrum of differential diagnosis surrounding it. Among these, primary TB of a skeletal muscle is the rarest form.^{3,4} Petter recorded a single case of primary muscular tuberculosis in 6,000 cases of all types of TB with a frequency of 0.015%.⁵ The skeletal muscles are very seldom affected by Tuberculosis as they do not serve a favourable environment for the growth, survival and rapid multiplication of mycobacterium tuberculosis.

CASE REPORT

A 15 year old unmarried female came to the surgery OPD with chief complain of swelling and pain over right forearm since 3 months and deformity of right hand since 2 months. Patient was apparently alright 3 months back when she noticed swelling over right forearm which was initially small in size and gradually progressed to present size. It was also associated with flexion deformity of right hand with inability to extend right little and right index finger. It was also associated with paraesthesia over right hand and forearm. Patient after experiencing these symptoms went to private hospital from where she got referred to the Surgery Dept. of St. George's Hospital, Mumbai for further management and evaluation. When evaluated here, we found that there was no history of prior trauma or surgery over the right hand or any intramuscular injection at the local site. She neither had any previous history of diabetes, immunosuppression nor any corticosteroid usage or renal failure in the past. She had no pyrexia, nocturnal sweats or any significant weight loss and peripheral stigmata of Tuberculosis were absent. There were no hypopigmented patches over her body and neither was there any h/o other medical/surgical illness in the past. On examination patient's vitals were found to be stable with a regular pulse of 80 beats per minute and with a BP of 120/80 mm of Hg. No signs of pallor, icterus, oedema and clubbing, were seen in the patient and she was afebrile. There was no axillary or cervical lymphadenopathy or any abscess seen. Systemic examination findings were unremarkable except for a 4x5 cm oval swelling over ulnar side of Right forearm, volar surface 5 cm in diameter. Swelling was diffuse, immobile, firm in consistency, non transilluminant and non-tender. No visible signs of inflammation over and around the swelling were seen. Similarly on examination of right hand flexion, claw deformity of RF, LF at PIP and DIP joints were supple. Loss of sensation over ulnar territory was predominant finding found which lead us to a provisional diagnosis of peripheral nerve sheath tumor of right ulnar nerve with claw hand. However, we were uncertain regarding our diagnosis and hence we decided to run few investigations.

Haematology/Biochemistry investigations revealed that the patient's ESR was markedly raised and was around 40mm/hr and patient's WBC count was 6000/cmm and her platelets, RBC counts were in normal range. Her renal and liver function test reports were also normal.

USG of Ulnar nerve revealed that the mass is well defined & hypo echoic with no signs of calcification or vascularity. The sonologist however suggested that the mass was way too hypo echoic than expected raising a suspicion over its neurogenic origin/schwannoma/neurofibroma and hence indicated it to be an atypical presentation emphasizing more on a histopathology report.

Nerve Conduction Studies revealed that the SNAP,

CMAP were within normal limits with no obvious signs of delay or impaired conduction in bilateral median and ulnar nerves with normal F wave latency and morphology. Similarly there were no signs of active denervation seen over right first dorsal interosseous, right flexor carpi ulnaris, right APB

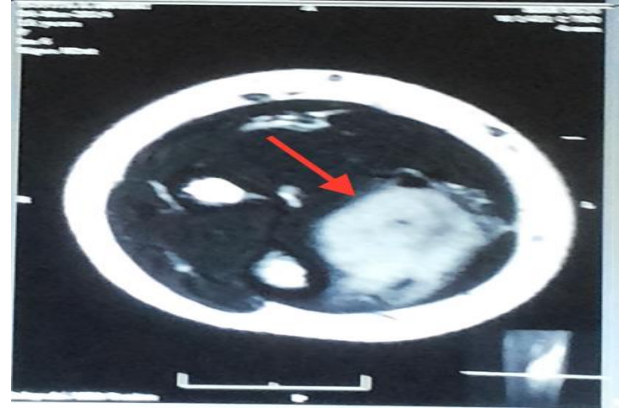


Figure 1: T2W (hyperintense).

MRI of the Right forearm was studied with 5 mm thin slices in axial and sagittal planes. T1W, T2W, PDFS, STIR SAG and MERGE images were obtained.

A well-defined solid spindle shaped focal lesion was seen on the ulnar aspect of mid forearm medially measuring 45 x 18 x 20 mm (length x AP x transverse). It was 'hypo intense' on 'T1W' and 'hyper intense' on 'STIR' and 'T2W'. Similarly no hemorrhage or calcification was noted within it. No obvious signs of any fracture, bone destruction or erosion were identified and the forearm muscles were found to be unremarkable and hence it became difficult to conclude the involvement of a specific forearm muscle. Bone marrow revealed normal signal. Radiologist came to a conclusion that it was a well-defined solid spindle shaped focal lesion on ulnar aspect of mid forearm medially mainly suggestive of peripheral nerve sheath schwannoma from ulnar nerve.

Because of a very higher suspicion of tumor and uncertainty surrounding the pathology and findings on investigations we decided to undertake exploration of the swelling/tumor and decided to perform excisional biopsy. Surgery was performed under general anesthesia. A linear incision was taken over the tumor site and incision was further deepened to reach the muscle. Ulnar nerve was identified and was found to be free from the tumor. Tumor was arising from a part of Flexor Digitorum Profundus (FDP). Caseous material was drained. All tumor mass with 1 cm margin was excised along with a part of FDP. Suction drain was placed in situ and incision was closed layer by layer. Patient withstood the surgery well. Excised swelling were handed over to scrub nurse for sending it for histopathological examination, which revealed that multiple large epithelioid cell granulomas with large central necrosis were seen and were comprised of large Epithelioid cells, Langerhans type Giant Cells

and Lymphocytes. Inflammation was seen extending into fibro fatty tissue and nerve bundles. Histopathologist's impression was that of a Granulomatous inflammation suggestive of tuberculosis of muscular tissue.

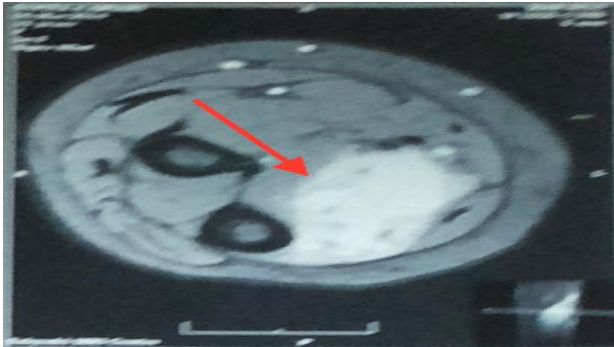


Figure 2: MERGE.

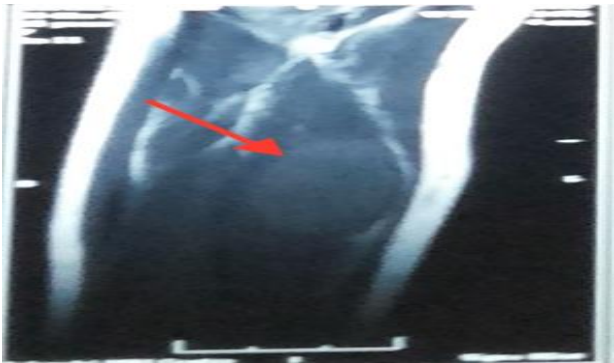


Figure 3: T1W (hypointense).

DISCUSSION

Incidence

Infection of *Mycobacterium Tuberculosis* is an extremely rare cause for pyomyositis. Predominantly seen manifestation is the pulmonary tuberculosis and tubercular lymphadenopathy. Although it is presumed to spread to the musculoskeletal system through a focus, the prevalence of active pulmonary tuberculosis coexisting with musculoskeletal TB has been 29% (147 of 499 patients).⁶ Primary skeletal muscle TB is extremely rare and earlier studies have reported only 4 cases of muscle tuberculosis in 2224 autopsy specimens from tuberculosis.⁷ In more recent studies about 3% of patients with tuberculosis have musculoskeletal involvement, mostly spondylitis, osteomyelitis or arthritis.⁸ It tends to mimic several malignancies and several inflammatory diseases leading to misdiagnosis. By far the commonest site of involvement is thigh with predominant involvement of quadriceps femoris.⁹ Few case reports in the literature about tubercular pyomyositis are in immunodeficient, HIV infected and renal failure patients or in patients on corticosteroids, immunosuppressive drugs or chemotherapy.^{9,10} It has also been described in immunocompetent patients within different muscles but

very rarely.^{11,12}

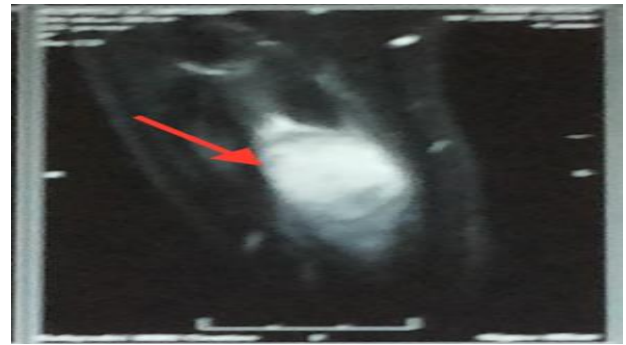


Figure 4: STIR SAG

Pathophysiology

In 63% of the cases tuberculosis spreads contagiously and in around 29 % of cases it spreads through the haematogenous route leaving 8% of infections to cause by direct inoculation. Besides this transmission by way of injection was also reported.⁴ The involvement of skeletal muscle in tuberculosis is usually by a direct extension from a neighbouring joint or by rarely by haematogenous spread. Skeletal muscles are known to be 'forbidden tissue' for growth and multiplication of tubercular bacilli.¹³ The pathophysiology is not well understood till today yet microbiologists and immunologists believe that because of poor oxygen content, high lactic acid concentration and paucity of reticuloendothelial cells and lymphatic tissue in muscles associated with very rich blood supply may help towards the localization of the bacteria in the muscles.¹⁴ Due to the inability of the mycobacteria to produce proteolytic enzymes they do not cause a pyogenic infection, but it may get secondarily infected leading to abscess formation in surrounding tissue.¹⁵



Figure 5: Excision of the tuberculous granuloma (intraoperative).

Tubercular pyomyositis can be attributed mainly to haematogenous dissemination of the tubercle bacilli and similarly it in the absence of a direct spread from an adjacent primary focus, it may be found in immunocompromised patients, inoculation through

needles and syringes contaminated with mycobacterium and idiopathically in an immunocompetent host.^{16,17}

Differential diagnosis

There is usually a delay in diagnosis because of the rarity and unusual/atypical presentation and similarly lack of knowledge, absence of early specific signs and a wide array of differentials associated with it. Actually the clinical manifestations are non-specific.

For a patient with pseudotumoural soft tissue lesions of hand and arm, there can be a wide array of differentials like anomalous and accessory muscles, anomalous osseous structures, tendon sheath cyst, synovial cyst, myositis ossificans, foreign body granuloma, epidermoid cyst, rheumatoid nodule, subcutaneous granuloma annulare, hypothenar hammer syndrome etc. Also metabolic disorders like gout, pseudo gout, amyloidosis also includes the differentials list.¹⁸ Tubercular pyomyositis is commonly misdiagnosed as soft tissue sarcoma, parasitic infection like cysticercosis or hydatid cyst (echinococcus), filarial infections as they also present as superficial palpable multiple nodular swellings in different parts of the body and inflammatory myositis or hematoma with secondary infection because of its close resemblance clinically.^{13,19} These diseases were ruled out by biomarkers, hematological indices, radiological investigations like MRI and also by using clinical features of the disease, lesion or infection. Similarly histopathological findings of caseating granuloma, absence of scolex of cysticercus, absence of cyst and fragments of acellular lamellate membrane of Echinococcus and absence of microfilaria including absence of eosinophils in the biopsy specimen of the patient.¹⁹ FNAC can prove a key in diagnosing parasitic infections and other but however a major quantum requires a biopsy for its correct diagnosis.¹⁹

Investigations

A raised ESR always points towards tubercular infections and is a consistent finding in these patients. Hence it must raise a suspicion in such patients for its diagnosis with intramuscular TB.

DNA-PCR is a highly sensitive investigation and helps in differentiating typical and atypical mycobacteria.

However contrast enhanced MRI is the investigation of choice and is superior to CT scan and USG in the detection and depiction of a swelling in order to differentiate it from malignancy.²⁰

Radiology-MRI

MRI presents with classical features of intramuscular TB with higher diagnostic accuracy. Especially on T1 weighted MRI low or intermediate signal intensity is obtained whereas in case of T2 weighted MRI high signal

intensity images of muscles are obtained. On gadolinium infusion a rim enhancement effect can be seen making the diagnosis even more clear.²¹ When T1 and T2 images are used, it becomes very difficult at times to differentiate neoplasms either benign or malignant from non-neoplastic disorders, including inflammatory pathologies. On T2 weighted MRI usually low intensity within the lesion represents debris and proteinaceous material.²² Fat suppressed STIR should be performed if there is doubt about the soft tissue inflammation secondary to infection or tumors.²³ As seen in our case on T1W MRI a hypo intense lesion was seen whereas the same lesion was hyper intense on T2W MRI. We also performed STIR sequence due to uncertainty surrounding the diagnosis of the lesion. However there are many more lesions, pathologies and disease processes, which produce similar muscle signal intensity on a MRI such as rhabdomyolysis, compartment syndrome, diabetic infarction, autoimmune and sarcoid myopathy or several infectious processes along with nerve sheath tumors and various other denervation disease processes and pathologies.²⁴ Hence biopsy or FNAC is the last resort when uncertainty regarding final diagnosis prevails.

Treatment

High suspicion along with ruling out various differentials is the key in diagnosing intramuscular TB. On diagnosis, ideally tuberculosis is treated with standard anti tubercular drug regimen including Isoniazid, Rifampicin, Pyrazinamide and Ethambutol. Usually treatment should be continued for either 6 months or 9 months. However owing to its rare occurrence and uncertainty on radiodiagnosis and wide array of differentials mimicking the disease usually surgeons prefer to perform excision and biopsy and after histopathology confirmation finally we can finally initiate AKT and treat patient. As in our case we followed this protocol and now patient is in good health without recurrence of the disease and without any secondary pathology elsewhere in the body.

Uniqueness of the case

In most of the cases published till today's date have mainly had the patients with immunocompromised status or with multiple site occurrence of the lesion and mostly involving thigh muscles. Our case is unique in the sense that the patient was immunocompetent and there was a simple solitary swelling over the right forearm muscle (FDP) without multiple site involvement and without any identifiable primary focus in the body in a tubercular endemic country like India.

CONCLUSION

Intramuscular tuberculosis is indeed a very rarely occurring phenomenon. The diagnosis can be made by high clinical suspicion and by ruling out various differentials and patient can be treated conservatively by initiating AKT regimen. Similarly one must focus on the

clinical features and radiodiagnosis. If the picture is still unclear and also if reaching to a proper diagnosis seems difficult then excisional biopsy and histopathology of the biopsy section can surely lead us to a correct diagnosis and further patient can be managed by pharmacotherapy and AKT.

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