

CASE REPORT

Diabetic Myonecrosis Involving Both Lower Limbs in Hemodialysis Patient: A Rare Complication of Diabetes

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Abstract:

Diabetic myonecrosis (DMN) is the term used for spontaneous ischemic necrosis of skeletal muscle, unrelated to atheroembolism or occlusion of major arteries. DMN is a rare microangiopathic disorder that can present as an acutely painful and swollen limb in patients with established diabetes mellitus. Muscles of the thighs are commonly affected in DMN. The condition can be diagnosed noninvasively with magnetic resonance imaging and resolves with analgesia, bed rest, and glycemic control. The majority of patients with DMN have diabetic nephropathy, yet this condition is not widely recognized. Due to uncommon presentation, lack of specific marker of investigation, diagnosis is often missed, resulting in unnecessary and deleterious interventions such as antibiotics, muscle biopsies, and surgery; the latter two of which can prolong recovery. Here, we report a patient on hemodialysis who is ultimately diagnosed as DMN affecting both lower limb and treated successfully.

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INTRODUCTION:

Diabetes mellitus is associated with microvascular and macrovascular complications like diabetic nephropathy, retinopathy and neuropathy. Less well-known complications are equally important, for diagnosis and treatment to decrease morbidity. Diabetic myonecrosis (DMN), also known as diabetic muscle infarction (DMI), causes spontaneous ischemic necrosis of skeletal muscle most commonly in the thigh or calf. DMN is usually unilateral and

affects the lower limbs. It was first described in 1965 by Angervall and Sterner as “tumoriform focal muscular degeneration”.¹ Concomitant diabetic nephropathy is present in 75% (95/126) of cases.² The pathophysiology is currently unclear, although thought to relate to microvascular dysfunction, with hypotheses including atheromatous occlusion, thrombus formation, endothelial damage, and dysfunction of local coagulation mechanisms.³ It presents clinically as sudden pain, swelling and tenderness of the involved muscle.⁴ Painful swelling may be acute or evolve over days to weeks. There is no specific marker for the disease. Diagnosis is based on clinical presentation, laboratory investigations and imaging, for which MRI is the modality of choice. Muscle biopsy should be reserved for atypical presentation, uncertain diagnosis, or when treatment fails to improve symptoms.

Bed rest, analgesia, and intense glycemic control are the cornerstones of diabetic myonecrosis therapy. It is important to note that a large majority have concomitant kidney disease with renal replacement therapy. With accurate and timely diagnosis and

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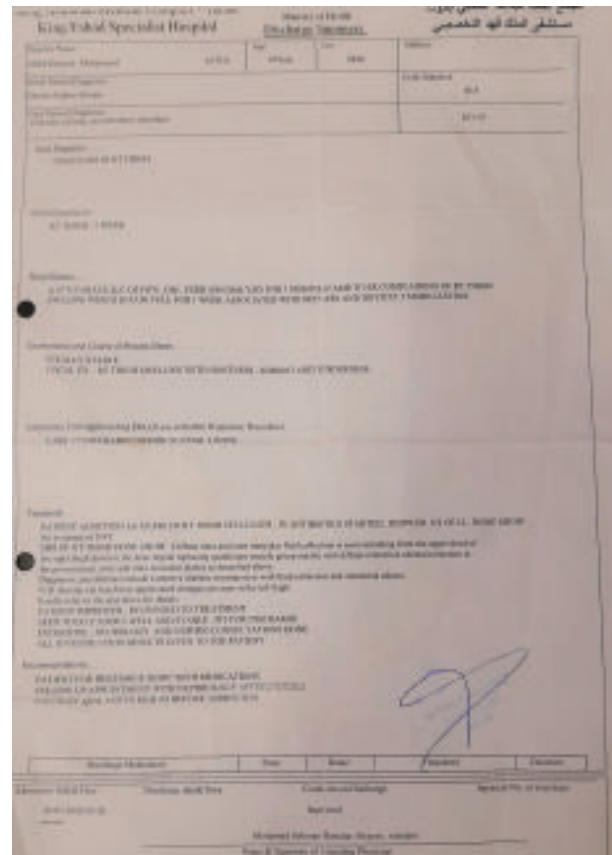
initiation of treatment, diabetic myonecrosis resolves spontaneously over a few weeks to months. Average recovery times were 5.5 weeks with aspirin and/or NSAID use, 8 weeks with bed rest and analgesics, and 13 weeks with surgical resection.¹⁰ Even with treatment, diabetic myonecrosis carries a high recurrence rate of 34.9 to 47.8% usually involving a contralateral limb within 6 months.^{4,11} Differential diagnoses include infection (pyomyositis, soft tissue abscess, osteomyelitis, cellulitis), tumors (lymphoma, sarcoma), and vascular pathologies (thromboses, compartment syndrome, calciphylaxis). Due to a relative lack of awareness regarding the condition, avoidable interventions such as muscle biopsies and even surgery are sometimes pursued, which have been associated with prolonged recovery times.^{4,12} A higher index of suspicion should be reserved for poorly controlled, longstanding diabetes patients with coexisting complications. Here we report a rare case of DMN affecting both thighs in patients on hemodialysis.

CASE SUMMARY:

A 45-year-old male presented to the emergency department (ED) with painful Rt. thigh swelling for 3 weeks. He has type 2 Diabetes Mellitus for more than 10 years, Hypertension for 1 year, End-stage renal disease (ESRD) on hemodialysis for 1 month. He complained redness and swelling of his rt. thigh and described his thigh pain as constant, aching, stabbing pain in the rt. posteriolateral mid-thigh with radiation distally to the calf. He denied any trauma or falls and reported worsening pain with weight-bearing and ambulation. With these complains, he admitted in King Fahad Specialist Hospital, Saudi Arabia on 30.07.22 and initially diagnosed as Cellulitis of Rt. thigh and treated with IV antibiotics and analgesics. X-ray of the right thigh, knee and lumbar spine, venous Doppler of the right lower extremity were normal. As improvement of his condition was not satisfactory, MRI of both thighs done. MRI of rt. thigh showed diffuse intra and inter muscular fluid collection seen extending from the upper third of the right thigh down to the knee region replacing quadriceps muscle group mainly with diffuse interstitial edema in the periosseous, inter and intra muscular plane and comment about possibilities of diabetes myonecrosis. MRI of lt. thigh showed similar yet much less appreciated changes. After 15 days with some improvement of rt. thigh, pt. complained of pain

in lt. thigh. The pt. discharged on 23.08.2022 and pt. returned to Bangladesh and admitted in Mugda medical college hospital.

On admission, he was afebrile with a pulse rate of 90 beats/min and blood pressure of 130/90 mm Hg. He had a swollen left thigh, tender to palpation. The overlying skin was palpably indurated with erythema and warmth with no evidence of discharge. Active and passive movements at the left hip and knee were limited due to pain. Lower extremity pulses were palpable bilaterally. Laboratory studies were remarkable for leukocytosis 20200/cumm, Hb 6.8 gm/dl, elevated creatinine kinase (CK) 436 U/L (42-196 U/L), C-reactive protein (CRP) 18 mg/dL (N<6 mg/dL), and erythrocyte sedimentation rate (ESR) 110 mm/hr (N 0-10 mm/hr). Additionally, poor glycemic control was confirmed with random blood glucose of 11 mmol/L and hemoglobin A1c 10.8%. Serology was negative for HIV and hepatitis B and C. Altogether, with these findings, previous MRI report and treatment history, a diagnosis of diabetic myonecrosis of lt. thigh was made. Patient condition improved with analgesics, with optimization of glycemia, regular thrice weekly hemodialysis via JVC.





DISCUSSION:

Diabetic myonecrosis is one of rare complication of diabetes. The first case of diabetic myonecrosis was described as 'tumoriform focal muscular degeneration' in 1965 by Angervall and Stener. They reported two patients, both underwent excision of a painful swelling in the proximal part of lower limbs, which showed ischaemic muscular necrosis.¹ Since then, around 200 cases have been described. It has been reported in both type 1 and type 2 diabetes. A systematic analysis of the available literature showed DMN was found in 54% in women, 50% had type 2 DM, mean age of presentation was 44.6 years, mean duration of diabetes at the time of DMN diagnosis was 18.9 years for T1DM and 11.0 years for T2DM. There was a preponderance of patients with other microvascular complications (65.8% had other microvascular complications) commonly renal failure (75% had nephropathy) reflecting poor glycaemic control, evident by mean HbA1c of 9.3%.⁴ Another study by Yong and Khoo analysed 24 publications with 41 patients having DMI.⁵ Of these, 53.7% were women, 41.5% had type 1 diabetes, 53.7% had type 2 diabetes, mean duration of DM at the time of DMI diagnosis was 17.3 years for type 1 diabetes and 15.7 years for type 2 diabetes, 29 patients were receiving renal replacement therapy in the form of haemodialysis (60.1%), peritoneal dialysis (21%) and renal transplant (12.2%) and the average HbA1c value was 7.2%, which is high considering the effect of renal failure on insulin metabolism. Our pt. was 45 years old male, suffered from Type 2 DM for 10 years, had ESRD on hemodialysis with uncontrolled DM (HbA1c 10.8%) which supports these data.

DMN is usually unilateral and affects the lower limb. The most commonly affected muscles are quadriceps, hip adductors, and hamstrings.⁶ Yong and Khoo in their study showed the muscle groups affected in diabetic ESRD patients on hemodialysis were thighs in 82% (22 cases), calves in 11% (3 cases), and upper limbs in 7% (2 cases) respectively.⁵ In our pt. both thigh muscles were affected. Bilateral limb involvement is uncommon.⁷ Bilateral involvement has been reported in 8.4% cases.⁹

Regarding diagnosis of diabetic myonecrosis routine laboratory investigations are relatively nonspecific. There may be leukocytosis and serum creatine kinase levels may remain normal or slightly elevated. MRI is the most sensitive diagnostic modality, and in the appropriate clinical setting muscle biopsy is not required. The characteristic MRI feature is an increased signal from the affected muscle area (intramuscular and perimuscular tissues), isointense or hypointense areas on T1-weighted images secondary to increased water content from edema and inflammatory changes that accompany the infarction.¹³ In our pt. there was leukocytosis, raised CK, ESR and CRP with characteristic MRI changes like diffuse intra and inter muscular fluid collection extending from the upper third of the right thigh down to the knee region replacing quadriceps muscle group mainly with diffuse interstitial edema in the periosseous, inter and intra muscular plane which supports the diagnosis of DMN.

The most accurate diagnostic modality is by tissue biopsy. On biopsy it grossly appears as nonhemorrhagic pale muscle tissue. Histologically, there are large areas of muscle necrosis and edema, phagocytosis of necrotic muscle fibres, granular tissue, and collagen.¹⁴ As muscle biopsies are more hazardous in HD patients as they got regular heparin during dialysis procedure, muscle biopsy was not done in our case. Muscle biopsy usually not done regularly in DMN as it is associated with poor wound healing and a higher risk of infection, seroma and haematoma. Therefore, currently it is recommended to make the diagnosis of DMN on the basis of clinical presentation and radiological findings; open muscle biopsy should be limited to atypical cases only.¹⁵

Treatment is conservative with supportive measures aimed at pain control with analgesics along with maintaining target glycemic control. Non-weight-bearing activity and physical rehabilitation may be

useful after the acute phase. Those who underwent surgery had an average recovery period of 13 weeks compared to 5.5 weeks for those only received conservative treatment¹¹. Our pt. was treated with regular thrice weekly hemodialysis with adequate control of DM (HbA1c 6.5% after a week of treatment), bed rest and paracetamol. 6 weeks after initial presentation, pt. condition improved markedly. Although diabetic myonecrosis has a good prognosis, it is an indicator of poor long-term outcome. However, recurrence rate are reported to be high (40%) with a 2 year mortality rate of 10%.¹⁶

CONCLUSION:

Diabetic myonecrosis is a rare and underreported complication of longstanding, poorly controlled diabetes mellitus. It can occur in diabetic renal patients with or without hemodialysis. Diagnosis requires clinically a higher index of suspicion for poorly controlled, longstanding diabetes patients with coexisting complications supported by MRI findings. Muscle biopsy is not routinely indicated and treatment is mainly conservative with analgesia and rest. However, the recurrence rate remains high, and long-term prognosis is poor. Awareness and early diagnosis of this condition will help to improve treatment, patient care, and prevent unnecessary invasive interventions or antibiotics.

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