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CASE REPORT

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Diabetic myonecrosis, an uncommon presentation of diabetes mellitus in tropical area: A case report

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Abstract

Diabetic myonecrosis is an uncommon complication related to long-standing poorly controlled diabetes. A 33-year-old Sudanese male patient with type one diabetes presented with progressive, severe bilateral thigh pain with low-grade fever. Laboratory results show hyperglycemia with ketonuria and elevated creatine kinase but normal white cell blood count. The patient was diagnosed initially with diabetic ketoacidosis with pyomyositis and received analgesic and insulin; the patient partially improved. After the second evaluation, bilateral thigh MRI was requested and shows diffuse edema involving the medial muscle group of the upper third of the right side with intramuscular facial edema, appearing as low signal in T1 and high signal in T2 and fat suppression images with no evidence of collection or abscess. Diagnosis of diabetic myonecrosis was made. The patient was managed conservatively and discharge on aspirin with full recovery.

KEYWORDS

diabetes, edema, muscle infarction, myonecrosis, tropical area

1 | INTRODUCTION

Diabetic myonecrosis is an uncommon complication of diabetes mellitus that occurs in patients with long-standing poorly controlled diabetes. Angervall and Stener first reported it in 1965 as focal muscular degeneration in 2 diabetic patients.[1]

Since then, less than 200 cases have been reported.[2] The pathogenesis of this rare entity is poorly understood. Literature reports the role of microangiopathy, atherosclerotic plaquing of microvessels, superimposed vasculitis, ischemia–reperfusion injury, and thrombosis of microvasculature, along with the causative role of coagulation fibrinolytic cascade with hypercoagulable state secondary to low antithrombin-III levels.[3–5]

The diagnosis of DMN is often very challenging and difficult. The diagnosis is often missed, or the condition is misdiagnosed unless the physician or radiologist is well aware of the clinical presentation and diagnostic clues.

Diabetic myonecrosis most commonly affects the thigh and usually presents with acute muscle pain, edema, and erythema in the absence of trauma or fever.[6]

Physical examination reveals swollen and tender muscle, mimicking deep venous thrombosis (DVT), and almost all the time presumptive diagnosis of DVT is made and often examination remains suboptimal due to fear of

throwing thrombus. However, if the examination is done properly, the muscle may feel indurated, which can suggest muscular etiology. At time, the diagnosis of lymphangitis/cellulitis is considered due to the presence of subcutaneous edema and fluid. Other differential diagnoses include abscess, necrotizing fasciitis, compartment syndrome, osteomyelitis, polymyositis, dermatomyositis, drug-induced myositis, and superficial thrombophlebitis. However, the absence of overlying erythema and classic constitutional symptoms should make physicians consider the diagnosis of DMN in long-standing poorly controlled diabetes with leg pain.[3]

Blood work classically shows normal to mildly increased WBC, normal to elevated inflammatory markers (ESR, CRP), and normal or mild elevation of muscle enzymes, like CPK.[5] Ultrasonography (US) is the initial imaging study that should be performed to rule out venous thrombosis, superficial thrombophlebitis, underlying abscess or the localized fluid collection, and necrotizing fasciitis.[7] Magnetic resonance imaging (MRI) is the next modality of choice with the sensitivity of T2-weighted MRI approaching 90% for picking up active muscle disease; however, specificity for muscle infarction is only 43%.[8]

2 | CASE PRESENTATION

A 33-year-old Sudanese male, self-employer who is known case of type one diabetes mellitus for 6 years, presented to an emergency department in Atbra hospital with bilateral thigh pain for 4 days with a low-grade fever for 2 days.

The pain was localized to the medial aspect of both thighs, gradual in onset which became very severe and interfered with his daily activity, increasing by movement and activity, decreasing by rest, and staying static. It is associated with generalized fatigability. There is no other group of muscles affected.

Fever was low-grade intermittent, not documented, and responded well to oral paracetamol.

The systematic review was unremarkable apart from poly-urea, nausea, and vomiting; urine was clear with normal color.

On reviewing his relevant past history, the patient was diabetic for 6 years on mixtard insulin 15-10 but poor control, less adherence to medications with frequent hospitalizations, no ICU admission, and no other chronic illnesses, no past history of upper respiratory tract infection, bloody diarrhea, or watery diarrhea. His family history was positive for D.M and negative for rheumatological disorder and no chronic medication apart from insulin.

The patient was in severe pain, febrile not documented by Atbra hospital staff, respiratory rate 30, and pulse rate 104 with regular radial pulsation. Peripheral lower limb pulsation was intact. There was severe tenderness on the medial aspect of both thighs with hotness and swelling. The patient cannot move his legs. Another neurological examination was normal with no upper limb or cranial nerve abnormality. No other complications of diabetes mellitus were detected.

TABLE 1 Investigations were done in the first evaluation in the hospital

Hb	13.5 g/dl
TWBC	9700×10^3
Random blood glucose	250 mg/dl
HbA1C	14%
Serum urea	42 mg/dl
Creatinine	0.7 mg/dl
Uric acid	4 mg/dl
Acetone (urine sample)	+++
Serum CK	>900 U/L
Serum calcium	7.2 mg/dl
Serum potassium	4.46 mEq/L
CRP	200 mg/L
ESR	25 at the first hour

The patient had high RBG and HbA1c. His inflammatory markers were raised with a normal white cell count. Serum creatine kinase (S. CK) was elevated (900 U/L) U&E, and liver enzymes were both within normal limits (Table 1). Doppler U/S was done and was negative for DVT.

The patient was diagnosed with diabetic ketoacidosis with polymyositis and admitted to the hospital for 6 days where he received I.V fluid, analgesia, insulin, and antibiotic; the patient recovered from DKA with a partial decrease in thigh pain and was discharged on Lantus 16 units with soluble insulin 8 units before each meal.

Then, the patient was referred to us in the rheumatology clinic at Haj Alsafi hospital. On second evaluation, the patient still had bilateral thigh pain with nausea and vomiting and was unable to walk and afebrile (37.6°). Serum creatine kinase level normalizes (25.6 U/L) and CRP level decreased, but still high (25 mg/L) CBC and LFT was all within normal.

A bilateral thigh MRI with I.V was done; it shows diffuse edema involving medial muscle group of the upper third of the right side with intramuscular facial edema, appearing as low signal in T1 and high signal in T2 and fat suppression images. The size of the affected right thigh is larger than the left. There is no enhancing abscess or free fluid collection, which is consistent with diabetic myonecrosis (Figure 1).

The diagnosis of diabetic myonecrosis was made, and the patient received analgesia. His blood glucose was optimized and underwent physiotherapy sessions. The pain gradually improved, and the patient was discharged on aspirin in good condition.

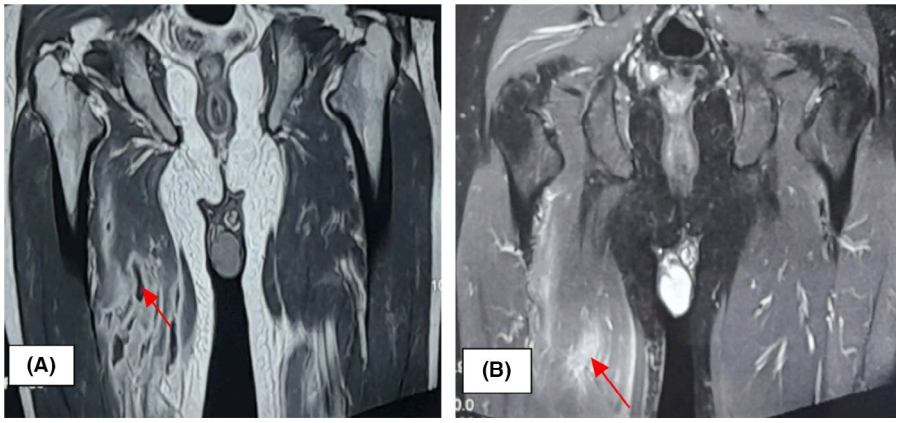

FIGURE 1 MRI of the thigh. (A) T1-weighted coronal view shows low signals. (B) T2-weighted coronal view shows high signals

3 | DISCUSSION

Diabetic myonecrosis (DMN) is a rare presentation in long-standing poorly controlled complicated diabetes, and it is often associated with misdiagnosis.[9,10] Although treatment is conservative, invasive procedure as biopsy and inappropriate management such as anticoagulation are associated with increased morbidity and delayed recovery.[10–12]

The most common affected muscle is the thigh muscle, which is consistent with our case.[11] Surprisingly as opposed to the classical age of presentation and diabetes status, our case was relatively young with diabetes onset of less than 15 years with no identifiable target organ complications. Although it is rare, bilateral involvement has been reported in 10% of cases.[10,11] The examination finding of hot, tender swollen muscle is a common finding and was elicited during the assessment. Some literature reports weakness and even sensory disturbance, and so our patient has a weak but intact sensation. Laboratory result of normal white cell count and mildly elevated CK with a poor glycemic index (RBG and HbA1C) was the same as in previous literature.[13,14]

Diagnosis of diabetic myonecrosis was challenging in this patient, as low-grade fever (although not documented) with DKA especially in tropical settings put the possibility of pyomyositis, which is a devastating disease that requires early surgical intervention. MRI is useful guidance for differentiation.

4 | LIMITATIONS

As the patient was admitted firstly to a low-resource area at Atbra hospital, and ABG was not available. So, DKA was diagnosed clinically with aid of high blood glucose and acetone in urine. In addition, no documented fever in the referral report from the peripheral hospital.

CONFLICT OF INTEREST

The authors report no conflicts of interest in this work.

AUTHOR CONTRIBUTIONS

Dr. Ziryab Imad Taha involved in writing paper, editing, diagnosis, and management. Dr. Yassin Abdelrahim Abdalla and Dr: Salih Boushra Hamza involved in writing paper, editing, and follow-up. Dr: Ali Ibrahim Elsiddig and Dr: Sami Ahmed Abdelgadir involved in reviewing images. Dr: Sohep Abdalla Osman involved in editing and follow-up.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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REFERENCES

1. Angervall L, Stener B. Tumoriform focal muscular degeneration in two diabetic patients. *Diabetologia*. 1965;1:39-42.
2. Trujillo-Santos AJ. Diabetic muscle infarction. *Diabetes Care*. 2003;26:211-215.
3. Mukhopadhyay P, Barai R, Philips CA, Ghosh J, Saha S. An unusual case of myonecrosis. *Case Rep Endocrinol*. 2011;2011:1-4.
4. Rastogi A, Bhadada S, Saikia U, Bhansali A. Recurrent diabetic myonecrosis: a rare complication of a common disease, Indian. *J Med Sci*. 2011;65(7):311.
5. Palmer GW, Greco TP. Diabetic thigh muscle infarction in association with antiphospholipid antibodies. *Semin Arthritis Rheum*. 2001;30(4):272-280.
6. World Health Organization. *Global Report on Diabetes*. World Health Organization; 2016.
7. Nagdev A, Murphy M, Sisson C. Bedside ultrasound for the detection of diabetic myonecrosis. *Am J Emerg Med*. 2008;26(8):969-e3.
8. Morcuende JA, Dobbs MB, Buckwalter JA, Crawford H. Diabetic muscle infarction. *Iowa Orthop J*. 2000;20:65.
9. Baker JC, Demertzis JL, Rhodes NG. Diabetic musculoskeletal complications and their imaging mimics. *Radiographics*. 2012;32(7):1959-1975.
10. Gupta S, Goyal P, Sharma P, Soin P, Kochar PS. Recurrent diabetic myonecrosis – an under-diagnosed cause of acute painful swollen limb in long standing diabetics. *Ann Med Surg*. 2018;35:141-145.

11. Stevens AC. Diabetic myonecrosis: a rare complication of diabetes mellitus mimicking deep vein thrombosis. *Am J Case Rep*. 2017;18:38-41.
12. Kapur S, Brunet JA, McKendry RJ. Diabetic muscle infarction: case report and review. *J Rheumatol*. 2004;31(1):190-194.
13. Storandt M, Thondapi C, Matta A. Case reports in diabetic myonecrosis: an uncommon complication of a common condition. *EJCRIM*. 2020;7(3):1389.
14. Jelinek JS, Murphey MD, Aboulafia AJ, et al. Muscle infarction in patients with diabetes mellitus: MR imaging findings. *Radiology*. 1999;211(1):241-247.

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