American Journal of Case Reports

e-ISSN 1941-5923
© Am J Case Rep, 2019; 20: 1268-1272
DOI: 10.12659/AJCR.917030

Received: 2019.04.17
Accepted: 2019.06.05
Published: 2019.08.28

Upper-Limb Diabetic Myonecrosis: Atypical Presentation of a Rare Complication

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Conflict of interest: None declared

Patient: Female, 55
Final Diagnosis: Diabetic ischemic myonecrosis of left arm (biceps muscle)
Symptoms: Erythema • pain upper arm • swelling
Medication: —
Clinical Procedure: Supportive therapy with analgesics • blood glucose control
Specialty: Rheumatology

Objective: Rare disease

Background: Myonecrosis is an uncommon complication of poorly controlled diabetes, predominantly involving the lower limbs. It is an atypical presentation in the upper limbs. Here, we report a rare case with atypical involvement of the upper limbs.

Case Report: A 53-year-old diabetic woman presented with left arm pain for the past week. She was not compliant with her medications. The patient denied any history of trauma or injection. Physical examination revealed a warm, tender, and erythematous swelling on the medial side of the left arm and was otherwise unremarkable. Her glycemic control was poor, with Hb A1C of 9.6%. Duplex ultrasonography demonstrated no evidence of fluid collection or thrombosis. An initial MRI (without contrast) report was misleadingly suggestive of polymyositis. Orthopedic consultant urged the patient to transfer to the operating room for aspiration of a probable infectious nidus, which resulted in a dry tap. Despite confusing radiological clues, ischemic myonecrosis was suspected, and second MRI studies (with contrast) reported necrosis. Tissue biopsy (the criterion standard) was withheld to avoid the risk of delayed healing or superimposed infection. Meanwhile, the patient received supportive treatment and achieved full recovery within 1 month.

Conclusions: Diabetic myonecrosis should be suspected in any poorly controlled diabetic patient presenting with otherwise unexplained muscle pain without any evidence of infection. Diagnosis can be made by MRI, leaving very few indications for invasive procedures. Analgesics and glycemic control are the mainstays of treatment.

MeSH Keywords: Arm Injuries • Diabetes Complications • Magnetic Resonance Imaging • Muscle, Skeletal • Necrosis

Full-text PDF: https://www.amjcaserep.com/abstract/index/idArt/917030

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Background

Myonecrosis, as one of the least expected complications of diabetes, typically manifests with extreme pain of the lower limb. Notwithstanding its complete resolution in the short term, the long-term prognosis is poor, with the majority of cases failing to survive 5 years following hospitalization for myonecrosis [1,2]. Although thigh muscles are the common sites of insult, there are rising numbers of case reports with upper-limb muscle involvement as well [2]. Herein, we present an atypical case of myonecrosis developing in the left arm of a patient known to have diabetes.

Case Report

A 53-year-old white (Turkic) woman with 20-year history of diabetes was referred to the rheumatology ward, tertiary hospital of Tabriz University of Medical Sciences, complaining of painful swelling on the medial side of left arm for the past week. Her other medical history was significant for ischemic cerebrovascular accident 3 years ago with no sequel and uncontrolled hypertension. She was reportedly noncompliant with her medications for glycemic control. Her last visit to the physician was 2 years ago; her most recent medication included daily glibenclamide 5 mg, insulin glargine 30 units, and daily captopril 50 mg. She had no history of tobacco smoking, alcohol, or substance abuse.

Physical examination revealed left arm swelling characterized by warmth, mild erythema, tenderness, and excruciating pain on motion (Figure 1). There was no evidence of gangrene. Peripheral pulses were symmetric and full. Muscle force and sensory exam of the affected limb were normal, although the patient resisted movement to avoid the pain.

She denied any history of prior trauma and injection at the site. The patient's temperature was 37°C, blood pressure was 170/100 mmHg, pulse was 76/minute, respirations were 16/minute, and oxygen saturation was 96% on room air. She appeared neither ill nor toxic. The list of differential diagnoses primarily consisted of thrombosis, cellulitis, fasciitis, abscess, pyomyositis, hematoma, muscle rupture, tumor, Fracture, and muscle infarction (myonecrosis). Laboratory and radiological imaging studies were conducted. On initial evaluations, soft-tissue edema was evident on plain radiography, without soft-tissue emphysema (Figure 2). The ultrasound revealed generalized muscular and subcutaneous edema with no collection or abnormal drainage in left arm and forearm.

Laboratory data were normal except for: random blood glucose=314 mg/dl (normal 79–160 mg/dl)/HbA1c=9.6% (normal

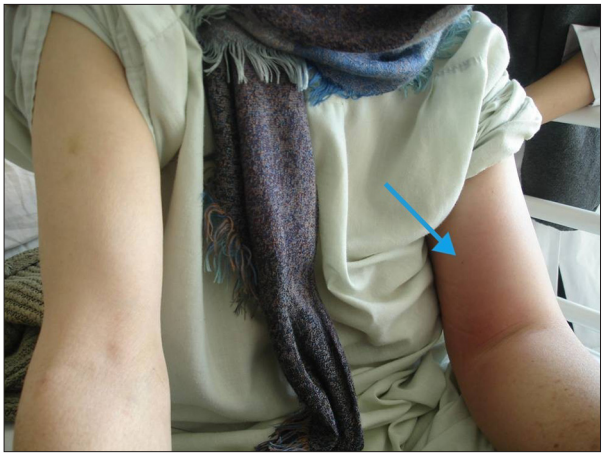

Figure 1. Ischemic myonecrosis of left arm in poorly controlled diabetic patient. She first presented with a painful left arm swelling. Erythema of overlying skin is notable compared to the right side (arrow).

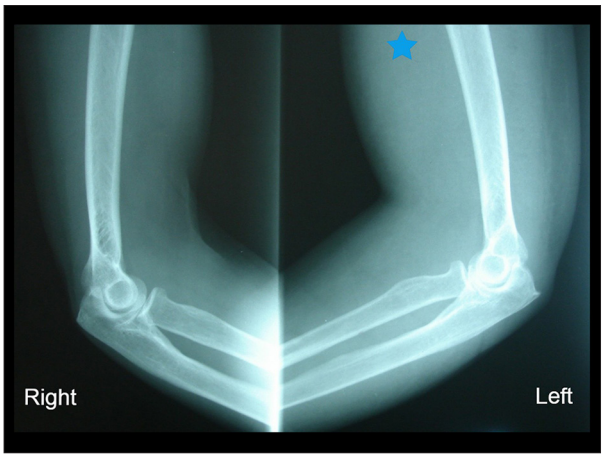

Figure 2. Upper limbs radiograph, lateral view. Significant soft-tissue swelling is present in the left arm, without any emphysema in the surrunding area (asterisks).

4–5.6%)/CRP= 4 mg/dl (normal <0.9 mg/dl)/CPK=229 IU/L (normal range: 24–170 IU/Liter) (Table 1).

In the absence of frank infection, the patient was referred for an MRI to evaluate for osteomyelitis. The report of MRI (without contrast) was as follows (Figure 3): "MR scan with axial and sagittal T1/T2/GE/W images are obtained. Abnormal high signal intensity is seen on biceps and all superficial and deep muscles of the forearm. Considerable subcutaneous edema is noted on the arm, elbow, and forearm. Bone marrow signal, joint spaces, and great arteries are normal. Superficial veins are collapsed. Triceps muscle seems to be intact. As a conclusion, the findings are compatible with arm and forearm polymyositis mainly in biceps."

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Table 1. Laboratory findings.

Blood, plasma, serum	Values	Reference range
Alanine aminotransferase	14 U/L	8–40 U/L
Alkaline phosphatase	187 U/L	45–115 U/L
Aspartate aminotransferase	13 U/L	8–40 U/L
Creatine kinase	229 U/L	10–70 U/L
Sodium	142 mEq/L	136–145 mEq/L
Potassium	4.5 mEq/L	3.5–5 mEq/L
Ferritin	53 ng/mL	12–150 ng/mL
Random blood glucose	314 mg/dL	<160 mg/dL
Serum iron	17 µg/dL	50–170 µg/dL
Lactate dehydrogenase	223 IU/L	100–250 IU/L
Blood urea	45 mg/dL	20–50 mg/dL
Creatinine	1.3 mg/dL	0.5–1.1 mg/dL
Erythrocyte sedimentation rate	77 mm/hr	0–20 mm/hr
Hematocrit	32.8%	36–46%
Hemoglobulin	10.2 g/dL	12–16 g/dL
Hemoglobin A1c	9.6%	<6%
Leukocyte count	7700	4500–11000/mm^3
Erythrocyte count	3.9 million/mm^3	3.5–5.5 million/mm^3
Mean corpuscular hemoglobin	26.2 pg/cell	25.4–34.6 pg/cell
Mean corpuscular hemoglobin concentration	31.3 Hb/cell	31–36% Hb/cell
Mean corpuscular volume	84.1 µm^3	80–100 µm^3
Platelet count	400,000/mm^3	150,000–400,000/mm^3
C-reactive protein	4 mg/dL	<0.9 mg/dL
Urine culture	Negative	–
Blood culture	Negative	–

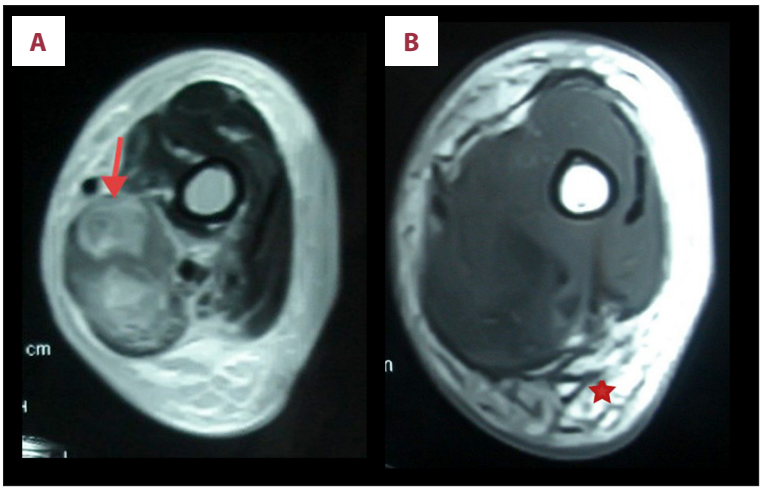

Figure 3. (**A, B**) First MRI without contrast. Left: Abnormal high signal intensity of biceps in T2 is shown (arrow). Bone marrow and triceps show normal signal. Right: There is remarkable subcutaneous and muscular edema in T1 (asterisk).

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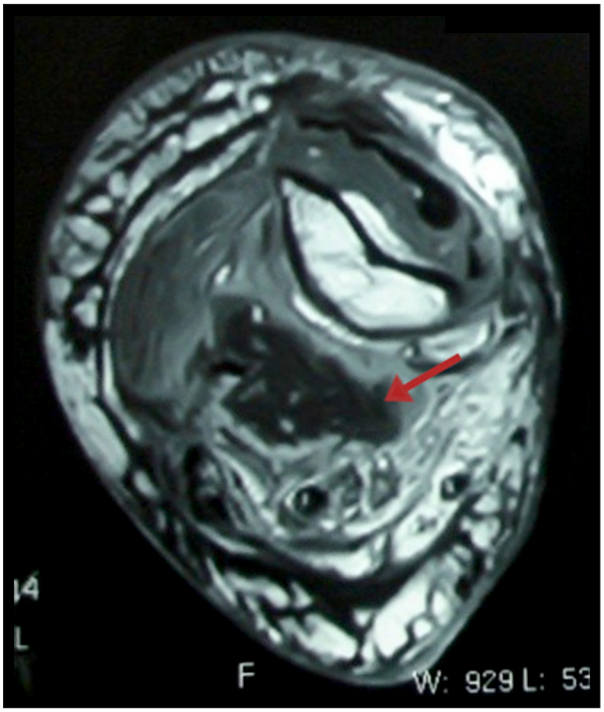

Figure 4. Second MRI (with contrast) conducted 2 weeks after the first MRI is significant for extensive edema centered on biceps, decreased signal intensity of biceps muscle on T1WI, edematous subcutaneous fat layers, and unremarkable bone marrow. Presence of nonhomogenous enhancement is noted after DTPA injection, which is suggestive of necrosis. Necrotic area in biceps muscle has failed to enhance with contrast (arrow).

Meanwhile, orthopedic consultation was performed, which led to a dry aspiration. A second sonography was requested, in which echogenicity of left biceps was remarkably enhanced and the muscle and subcutaneous fat layer were edematous, but no collections could be found. The biceps muscle showed hyperemia as well. Based on sonographic findings, hematoma and fluid collections were ruled out, and the radiologist suspected space-occupying lesions or inflammatory myositis of the biceps muscle. The patient's clinical condition showed little change. A second MRI was performed 2 weeks after first MRI, this time with contrast, and the report was as follows (Figure 4): "The examination was performed using post-contrast injection, which reveals the decreased signal intensity of biceps muscle on T1WI. Nonhomogenous enhancement was noted after DTPA (diethylenetriamine penta-acetic acid) injection. Some areas were not enhanced, probably due to necrosis. Subcutaneous fat layers were also edematous, while bone marrow was unremarkable." Despite partially confusing radiologic and orthopedic consultations, misdiagnosing the case with inflammatory or infectious myositis, we firmly withheld antibiotic therapy in the absence of any findings supporting an infectious process. We also ruled in the diagnosis of myonecrosis based on final MRI findings and avoided an invasive biopsy. As a result of supportive treatment with analgesics and glycemic control, swelling of the arm improved within a month, as expected of pathologically proven cases of myonecrosis.

Discussion

Ischemic myonecrosis is a remarkable yet overlooked diagnosis and mostly affects poorly controlled diabetics. It was initially introduced in 1965 [1]. Myonecrosis needs to be high on the list of differential diagnoses offered for any diabetic patient with spontaneous painful swelling of limbs [1]. The definitive diagnosis of non-infectious myonecrosis is often challenging, and many patients first require multiple testing to exclude more common differentials [2,3]. Despite its controversial pathophysiology, atheroembolism being superimposed on a diabetic's already diseased small vessels is the one mostly argued about [2]. However, no atheromatous plaques have been detected on histopathology slides obtained from living tissues or postmortem examinations [1]. While the contribution of arteriosclerosis obliterans to ischemic myonecrosis is one of the widely agreed upon underlying pathologies, development of compartment syndrome is also thought to exacerbate the existing ischemia [2]. Clotting cascade or fibrinolytic pathway abnormalities be partly responsible, but no substantial evidence has been found to support [2,4].

In contrast to our case, the typical clinical presentation of myonecrosis is thought to involve the medial aspect of the anterior thigh, followed by the medial compartment of the thigh and hamstrings [1]. Unlike our case, history of narcotics use, fever, trauma, or bedridden states are usually present [2,5]. MRI is by far the best modality used for the diagnostic evaluation of myonecrosis, as it is sensitive and offers the additional advantage of noninvasiveness [2]. Display of high-intensity signals on T2 and notable muscular edema extending to the adjacent hypodermal fat and peripheral connective tissue are characteristic findings on imaging [1,6].

Some physicians take the risk of delayed healing and secondarily imposed infections into account and argue for the adequacy of MRI, calling for bypassing the so-called criterion standard of biopsy unless evidence of other diagnosis is found [2]. However, biopsy of the involved tissue is practiced by some physicians, so that necrosis and arteriosclerosis can be viewed [2]. In the absence of a solid consensus regarding myonecrosis on the one hand, and the profoundly infrequent clinical encounters with the disease on the other hand, clinical management of diabetic myonecrosis used to be a matter of debate [2]. Our case is consistent with the bulk of studies in support of believing in MRI's adequacy for diagnosis, especially in the absence of evidence of ongoing infectious or inflammatory processes [1].

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Myonecrosis represents an abysmal control of diabetes. Establishing the diagnosis of myonecrosis is firm evidence of how severe the underlying vascular disease has already become, and these patients, independent of the course of myonecrosis, have an increased vulnerability to serious complications of micro- and macrovascular nature [7]. Multiple studies assessing the natural history of patients with diabetic myonecrosis have found that ischemic necrosis is a strong predictor of future cardiovascular morbidity and mortality [1,8]. Patients with complete resolution of myonecrosis have an increased risk of both fatal and nonfatal myocardial infarction and stroke and an elevated risk of death due to cardiovascular causes [6,8]. The occurrence of diabetic myonecrosis could be a cardiovascular disease risk equivalent, and these patients may benefit from aggressive risk factor modification for prevention of associated macrovascular causes of morbidity and mortality [1,2,9]. The treatment for non-infectious myonecrosis is supportive. However, patients whose glycemic control is poor enough to result in ischemic myonecrosis require extensive workup of the vital organs supplied by a hitherto impaired circulatory system [2,9]. If it remains unattended, they are prone to sustain further multiple organ injuries [2,9]. Ischemic myonecrosis and vascular complications of diabetes are thought to share some similar mechanisms, which is why patients are required to undergo thorough evaluations for end-organ damages promptly after the resolution of myonecrosis [1,10]. Unfortunately, our case was not an exception to the rule, and despite satisfactory resolution of myonecrosis in response to supportive therapy with analgesics, the ultimate result was dramatic. Aggressive glycemic control to delay life-threatening complications of underlying diabetes was of little success. She sustained a fatal myocardial infarction 1 year after her discharge, in a course similar to that of most of patients with myonecrosis, who die after discharge due to the complications of long-standing diabetes [2].

Conclusions

The occurrence of diabetic myonecrosis is an ominous indicator of the severity of underlying vasculopathy. It needs prompt attention, and if left unaddressed can lead to a devastating outcome.

Owing to the unfamiliar nature of diabetic myonecrosis to our radiologist and orthopedic consultants in charge of the case, we had to overcome the hurdle of taking diagnostic steps, without performing unnecessarily invasive procedures. It is recommended for physicians to expand their knowledge regarding such a reasonably rare but immensely serious complication, especially in the noncompliant diabetic population.

Acknowledgments

This research was generally supported by the Department of Rheumatology of Tabriz Medical Sciences. We thank our colleagues from Sina Hospital and Imam Reza Hospital who provided insight and expertise that greatly assisted in patient management.

We thank Dr. Mohammad Hossein Daghighi, radiologist, Imam Reza Hospital for his comments and timely consult throughout the process of attempting to making the correct diagnosis.

We would also thank Dr. Jafar Ganjpour, orthopedic surgeon, Shohada Hospital of Tabriz for sharing his wisdom with us in the course of treatment, although any errors are our own and should not tarnish the reputations of these esteemed persons.

Finally, it is unfortunate that our patient could not survive the consequences of her poorly controlled diabetes long after her hospital discharge.

Conflict of interest

None.

References:

1. Wintz RL, Pimstone KR, Nelson SD: Detection of diabetic myonecrosis: Complication is often-missed, sign of underlying disease. Postgrad Med, 2006; 119(3): 66–69
2. Horton WB, Taylor JS, Ragland TJ, Subauste AR: Diabetic muscle infarction: A systematic review. BMJ Open Diabetes Res Care, 2015; 3(1): e000082
3. Choudhury BK, Saikia UK, Sarma D et al: Diabetic myonecrosis: An underreported complication of diabetes mellitus. Indian J Endocrinol Metab, 2011; 15(Suppl. 1): S58
4. Mukhopadhyay P, Barai R, Philips CA et al: An unusual case of myonecrosis. Case Rep Endocrinol, 2011; 2011: 624020
5. Larson RC, Sierra RJ, Sundaram M et al: Calcific myonecrosis: A unique presentation in the upper extremity. Skeletal Radiol, 2004; 33(5): 306–9
6. Joshi R, Reen B, Sheehan H: Upper extremity diabetic muscle infarction in three patients with end-stage renal disease: A case series and review. J Clin Rheumatol, 2009; 15(2): 81–84
7. Lawrence L, Tovar-Camargo O, Lansang MC et al: A diagnostic and treatment challenge in longstanding diabetes. Case Rep Endocrinol, 2018; 2018: 1723695
8. Habib GS, Nashashibi M, Saliba W, Haj S: Diabetic muscular infarction: Emphasis on pathogenesis. Clin Rheumatol, 2003; 22(6): 450–51
9. Kapur S, Brunet JA, McKendry RJ: Diabetic muscle infarction: Case report and review. J Rheumatol, 2004; 31(1): 190–94
10. Galeano-Valle F, Benito-Martinez E, Alvarez-Sala-Walther L et al: An atypical presentation of diabetic myonecrosis. AACE Clini Case Rep, 2019; 5(1): e77–81

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