A Convoluted Picture of Diabetic Myonecrosis

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ABSTRACT

Introduction: A patient with well-controlled type 2 diabetes was found to have diabetic myonecrosis, a rare condition associated with poorly controlled type 2 diabetes. Diagnosis was masked by concern for lumbosacral plexopathy from a history of spinal cord infarct.

Case Presentation: A 49-year-old African American woman with type 2 diabetes and paraplegia secondary to spinal cord infarct presented to the emergency department with left leg swelling and weakness from her hip to toes. Hemoglobin A1c was 6.0%, and there was no leukocytosis or elevated inflammatory markers. Computed tomography showed evidence of infectious process or possible diabetic myonecrosis.

Discussion: Recent reviews show fewer than 200 reports of diabetic myonecrosis since first described in 1965. It typically is seen in poorly controlled types 1 and 2 diabetes, with average hemoglobin A1c of 9.34% at time of diagnosis.

Conclusions: Diabetic myonecrosis should be considered in diabetic patients with unexplained swelling and pain – particularly in the thigh – even with unremarkable lab values.

INTRODUCTION

Diabetic myonecrosis is a rare condition associated with poorly controlled type 1 and type 2 diabetes.[1] There have been few reported cases in the literature over the past several decades, despite the increasing worldwide prevalence of type 1 and type 2 diabetes.[1] Our case presents a patient with relatively well-controlled type 2 diabetes found to have diabetic myonecrosis, who presented with lower extremity weakness and edema that initially was confounded by concern for lumbosacral plexopathy due to her history of spinal cord infarct.

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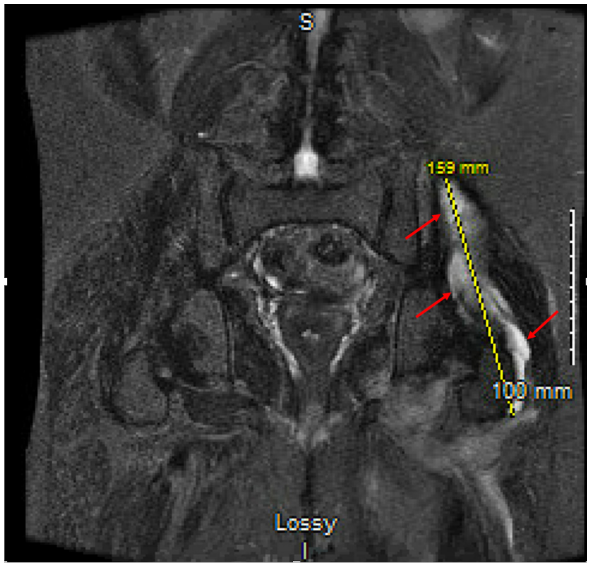
CASE PRESENTATION

A 49-year-old African American woman with a history of type 2 diabetes, hypertension, hyperlipidemia, and incomplete paraplegia secondary to spinal cord infarct complicated by neurogenic bowel and bladder presented to the emergency department with left leg swelling. Approximately 1 month prior to admission, she noted significant weakness in her left leg from her hip to her toes increased from baseline, without deficits in the right leg. There was swelling and a tingling sensation that did not improve with gabapentin or baclofen. She denied any systemic symptoms or new numbness in the leg. She has a history of a ventral spinal cord infarct in a T2-T8 distribution with decreased pinprick and temperature sensation at the T8 level, as well as weakness and absent patellar/ankle jerk reflexes in the bilateral lower extremities. She has spastic paraparesis for which she receives botulinum toxin injections, though there is no evidence of kidney disease secondary to her neurogenic bladder. Because of this history, there was concern for lumbosacral plexopathy; however magnetic resonance imaging (MRI), x-ray, and duplex ultrasound of the spine at this admission were unremarkable.

Admission lab work did not show leukocytosis or elevated inflammatory markers, reducing suspicion for an infectious or inflammatory cause. Ultrasound of the left lower extremity was negative for deep vein thrombosis (DVT), but computed tomography of the abdomen/pelvis showed low-attenuation areas in the left hip tissues concerning for an infectious process or diabetic myonecrosis. MRI demonstrated diffuse edema-like signal centered in the adductor compartment of the left thigh

Figure. Magnetic Resonance Imaging of the Pelvis With and Without Contrast



Red arrows pointing to areas of edema-like signal in adductor compartment, yellow line demonstrating length of likely muscle infarct.

with multiple areas of nonenhancement on postcontrast imaging, consistent with diabetic myonecrosis (Figure).

The patient was continued on aspirin therapy. With help from physical and occupational therapy, she endorsed improvement of weakness and symptoms during the hospital stay. She was discharged home on day 5 with outpatient physical therapy follow-up. Three months later, a follow-up electromyography study demonstrated lower motor neuron denervation changes in a diffuse yet patchy distribution consistent with probable diabetic myonecrosis in the left lower extremity. Together these results support diabetic myonecrosis as the cause of muscle infarct over other causes, such as the prior spinal cord infarct.

The patient was diagnosed initially with type 2 diabetes 7 years ago (2015), and her glycemic control is currently managed with metformin alone. Her hemoglobin A_{1c} (HbA_{1c}) value was 6.0% on this admission and 7.2% eight months ago. Her recent diabetic eye exam did not show evidence of retinopathy; her creatinine levels have remained stable and low for the past 4 years; and there was no evidence of diabetic neuropathy on exam. This suggests that her type 2 diabetes was well controlled at the time of diagnosis. She did mention that upon initial diagnosis of diabetes, she was started on insulin therapy and had a HbA_{1c} greater than 10%.

DISCUSSION

Diabetic myonecrosis is a rare condition associated with poorly controlled diabetes.[1] Recent reviews have shown fewer than 200 reports of this condition since it was first described in 1965.[1] It is typically seen in individuals with advanced diabetetes—both type 1 and type 2.[1] A recent case review demonstrated a mean reported HbA_{1c} of 9.34% at the time of diagnosis.[1] The pathophysiology is unclear, but proposed mechanisms include injury secondary to atherosclerosis, diabetic microangiopathy, vasculitis with thrombosis, or ischemia reperfusion injury.[2]

Suspicion for this rare condition should be increased in patients with poorly controlled diabetes presenting with acute muscle pain/swelling, particularly in the lower extremities. It is important to rule out acute conditions, such as DVT and trauma, in these patients as well, as they can sometimes present similarly or be related to underlying conditions such as sickle cell disease, which is more prevalent in African American populations.[3] In this patient without a history of sickle cell disease, the confounding factor was the previous spinal cord infarct that masked the picture, demonstrating once again how this condition is not routinely suspected. Routine laboratory values typically are not elevated, and blood and urine cultures typically do not yield any significant results.[1] However, inflammatory markers, such as erythrocyte sedimentation rate and C-reactive protein, appear to be somewhat useful and elevated in a large proportion of previous cases in which they were reported.[1] This patient's lab work, including inflammatory markers, was unremarkable and did not contribute to the diagnosis.

Typical locations of pain in these patients are in the lower extremities, particularly in the thighs.[1] This was seen in our case as well, with significant left thigh pain, swelling, and tingling sensations. Muscle biopsy can provide a definitive diagnosis; however, typically it is not done as it roughly doubles the time to symptom improvement.[1,4] MRI is sensitive and specific enough to diagnose diabetic myonecrosis without biopsy, showing edema with T2 hyperintensity or T1 iso-intensity or hypointensity.[1]

Therapies for diabetic myonecrosis should be targeted towards reduced recovery time and reduced rates of recurrence, as patients with diabetic myonecrosis are at high risk for recurrence of the condition.[1] The most effective evidence-based management for diabetic myonecrosis includes rest, analgesia using nonsteroidal anti-inflammatory drugs, and strict glycemic control.[1] Interestingly, the patient presented here had a recent HbA_{1c} of 6.0%, suggesting good glycemic control. Low-dose aspirin has been shown to reduce recovery time from 57 days to 39 days, on average, and reduce recurrence rates to 10% versus 32% for those on bed rest alone.[1] Surgical intervention has been shown to increase recovery time to 91 days and increase the recurrence rate to 50%.[1,5] While patients receiving physical therapy had a prolonged recovery time, the recurrence rate was reduced compared to those who were put on bed rest alone (18% vs 32% recurrence).[1] Because of this, physical therapy should be avoided in the acute phase of the illness but is recommended once patients are discharged from the hospital.

CONCLUSIONS

This case represents an even more rare case of diabetic myonecrosis in the setting of well-controlled type 2 diabetes, although the

patient's glycemic control was poor in the past. This finding suggests that diabetic myonecrosis should be considered in patients with diabetes presenting with extremity swelling, even in those with improved glycemic control. The presentation of this patient in the setting of recent spinal cord infarct, as well as normal lab markers and currently well-controlled type 2 diabetes, may serve to assist in making the diagnosis of diabetic myonecrosis in patients with an otherwise noncontributory workup and well-controlled type 2 diabetes.

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