

Case report

Diabetic myonecrosis: An unusual complication of Type 1 Diabetes Mellitus in a 49-year-old male patient. Case report.

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ABSTRACT

Diabetic myonecrosis (DMN) is an uncommon complication of uncontrolled Diabetes Mellitus (DM), resulting from microvascular changes that lead to ischemia and tissue necrosis. The objective of this study is to describe the clinical manifestations, diagnosis, and treatment of DMN through a case report. A 49-year-old male patient with poorly controlled type 1 DM with a 28-year history of the disease, presented with pain and swelling in his left thigh. Ultrasound revealed inflammation of the quadriceps femoris, suggesting pyomyositis. However, after two months without improvement despite multiple antibiotic regimens, magnetic resonance imaging (MRI) was ordered. The MRI showed no abscesses but rather a large inflammatory process with interstitial fluid visualized as hyperintense on T2-weighted images, establishing the diagnosis of DMN. He was treated with anti-inflammatory drugs, analgesics, glycemic control, and physical therapy. There was an initial favorable clinical course; however, he died eight months later from complications of distal penile necrosis and sepsis. DMN is more prevalent in women; it is unilateral in most cases, and it is often accompanied by microvascular complications. Clinical suspicion and magnetic resonance imaging (MRI) lead to an accurate diagnosis. Treatment focuses on analgesia, glycemic control, and rest. The short-term prognosis is favorable, although life expectancy does not exceed five years due to the high incidence of recurrence and death from vascular complications. Physicians should be made aware of this unusual condition and adopt a multidisciplinary approach for timely diagnosis and treatment.

Keywords: Diabetes Mellitus, diabetic myonecrosis, magnetic resonance imaging, quadriceps muscle.



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Introduction

Diabetes Mellitus (DM) is an endocrine and metabolic disease that currently constitutes one of the main health issues worldwide. Its high prevalence estimates that around 425 million people, or 8.8% of adults aged from 20 to 79 years old, suffer from it [1]. This disorder is characterized by hyperglycemia resulting from altered insulin production, decreased sensitivity in peripheral receptors, or both conditions. In the long term, this is associated with damage to various tissues and organs, including neuropathy, nephropathy, and diabetic retinopathy as widely recognized complications, as well as an increase in musculoskeletal system disorders by altering connective tissue components and thus causing chronic damage [2].

Diabetic myonecrosis (DMN), also known as ischemic myositis, is an infrequent complication associated with patients with uncontrolled type 1 and type 2 DM. It was first described in 1965, since then fewer than 200 cases have been reported worldwide. It originates from microangiopathic changes leading to muscular ischemia and subsequent tissue infarction. It occurs more frequently in females and alongside other diabetes-associated complications, the most common being diabetic nephropathy [3,4]. The clinical picture is characterized by severe and acute pain localized in the lower extremities, most frequently affecting the quadriceps femoris. The most common sign is unilateral swelling, which may or may not be accompanied by fever and mild leukocytosis [4].

Magnetic resonance imaging (MRI) is the diagnostic gold standard, typically revealing muscle edema in T2-weighted sequences due to their sensitivity to fluid. Likewise, a biopsy can be performed if other differential diagnoses such as pyomyositis are suspected; however, tissue damage from the sample collection combined with poor vascularity can lead to complications that delay the recovery process [4]. Optimal treatment is conservative, based on nonsteroidal anti-inflammatory drugs (NSAIDs) and strict rest [3]. The objective of this research

is to describe the clinical manifestations, diagnosis, and treatment of diabetic myonecrosis as an unusual complication of type 1 Diabetes Mellitus through a case report.

Case presentation

A 49-year-old male patient, diagnosed with type 1 Diabetes at the age of 21, treated irregularly with glargine insulin, presented with a three-month history of high-intensity, sudden-onset, non-remitting pain localized in the left thigh with no history of previous trauma. He also presented with swelling in this segment, beginning on the anterior aspect and extending to the posterior and medial aspects, which progressively worsened and limited ambulation.

He reported an allergy to NSAIDs, decreased visual acuity associated with grade 3 diabetic retinopathy diagnosed 10 years ago, and stage 5 chronic kidney disease (CKD) on hemodialysis for the last 8 years. He reported a history of Wagner grade II diabetic foot, two months prior to his current illness, treated with antibiotic therapy and local wound care, which evolved satisfactorily within a month. Additionally, he has a history of long-standing peripheral diabetic neuropathy treated with 1200 mg of gabapentin daily.

Physical examination revealed swelling on the left thigh encompassing its anterior, medial, and posterior aspects, extending from the groin to the knee, with a hardened area on the anterior and inner aspects of this zone, with tenderness but without color changes, pitting edema or temperature changes. Proximal range of motion was limited by pain, capillary refill time was less than 3 seconds, with palpable, symmetric, low-amplitude pulses. Distal sensation and mobility were preserved and symmetric compared to the contralateral side.

Initial laboratory reports showed HbA1c of 11.8% and fasting blood glucose of 310 mg/dL. C-reactive protein (CRP) 68 mg/L (reference < 5 mg/L), total creatine kinase (total CK) 480 U/L

(reference < 200 U/L), leukocytes 11,800/mm³ with 78% neutrophils, urea 112 mg/dL, and serum creatinine 6.4 mg/dL.

A skin and soft tissue ultrasound of the left thigh was requested, reporting an inflammatory process of the quadriceps femoris and suggesting pyomyositis, thus, empirical antibiotic therapy was initiated; however, there was no clinical improvement after two months of multiple antibiotic regimens. He sought medical attention once again, at which point an MRI was ordered. The MRI showed no collections or abscesses, but rather a large inflammatory process with free interstitial fluid visualized as hyperintense on T2-weighted images, (Figures 1 and 2). Given this finding and following discussion with a multidisciplinary team, the diagnosis of diabetic myonecrosis was established.

A muscle biopsy was considered to confirm the diagnosis, but it was contraindicated due to high risk of complications. Given the severity of the inflammatory process and quadriceps pain, and the impossibility of using NSAIDs, low-dose deflazacort (6 mg/day) was prescribed along with tramadol/paracetamol (75/650 mg every 8 hours) and clopidogrel (75 mg/day). The glucocorticoid use was limited to a short 3-week course, accompanied by rehabilitation therapy to prevent contractures and strict glycemic control due to high recurrence rate. NSAIDs were not prescribed due to a double contraindication: a history of allergy and the presence of stage 5 CKD. The clinical course was favorable, with improvement in pain and induration within three weeks. However, eight months later he returned to consultation presenting necrosis on the distal portion of the penis, which within a month led to local infection, sepsis, and ultimately the patient's death.

Figure 1. Simple left thigh magnetic resonance imaging. T1-weighted and T2-weighted images. Coronal section of the left femur, T1 image (a). Coronal section of the left femur, T2 image (b). Source: Medical records.

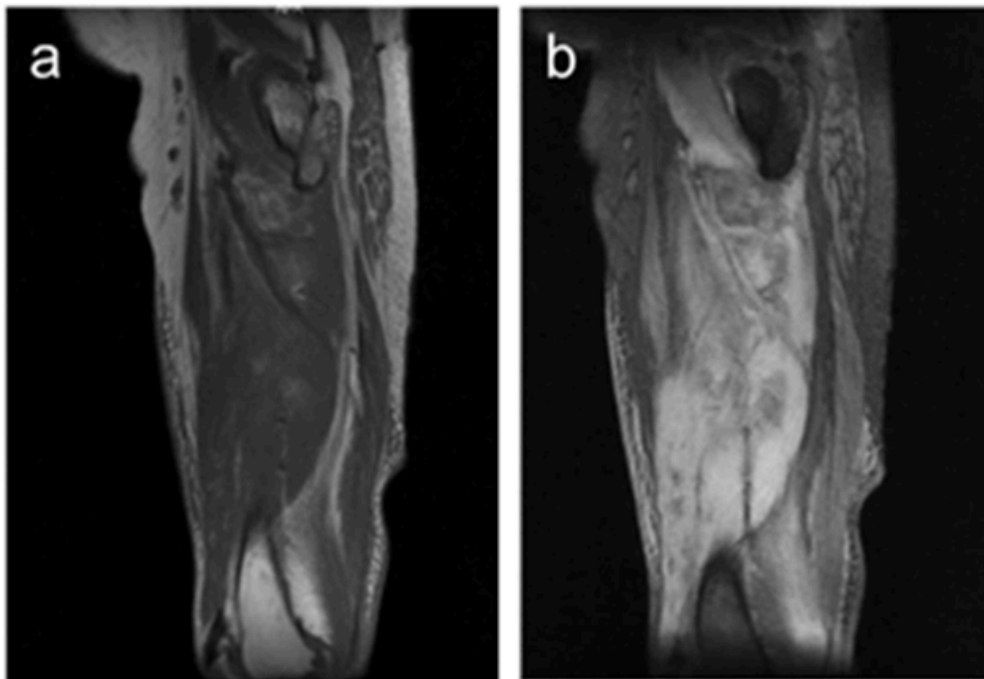
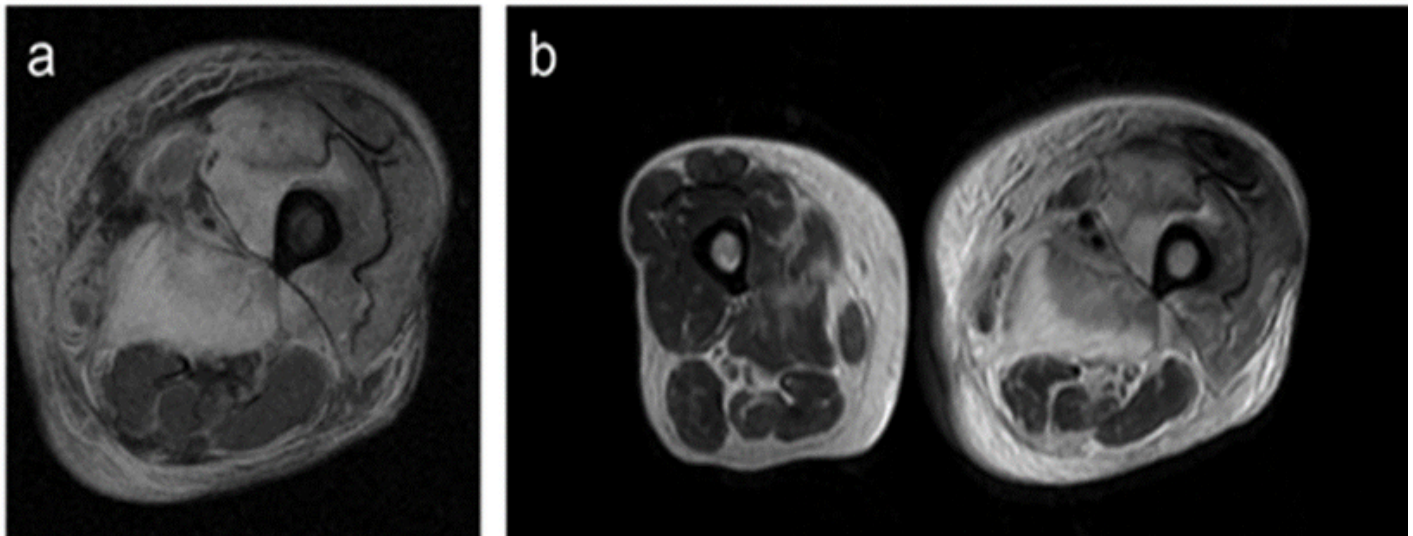


Figure 2. Simple left thigh magnetic resonance imaging. T2-weighted image. Axial section of the left femur (a). Axial section of both lower limbs (b). Source: Medical records.



Discussion

Diabetic myonecrosis is a complication that can occur in patients with either type 1 or type 2 diabetes, with prolonged disease duration and inadequate glycemic control being critical aspects that increase the risk of developing it [5]; nevertheless, some authors suggest it is more frequently associated with type 2 DM [6]. It is established that the average duration of prior underlying pathology at diagnosis is approximately 19 years for type 1 DM and 11 years for type 2 DM, presenting with a slightly higher prevalence in women with a mean age of 44.6 years [7]. This contrasts with this case, which centered on a male type 1 diabetic patient with irregular treatment and a much longer clinical course, which is a relevant factor in the development of this condition.

It is described that this entity presents concurrently with retinopathy, nephropathy, or neuropathy in 46.6% of cases, and that 65% of cases present at least two of these complications [3]. This was also found in the present case, as the patient presented all these microvascular complications, in addition to a history of grade II diabetic foot prior to the onset of myonecrosis, despite being older than average.

In the pathophysiology of DMN, microvascular occlusion stands out as a key factor leading to tissue necrosis [8]. Furthermore, some authors detail that it is secondary to atherosclerosis, diabetic microangiopathy, vasculitis with thrombosis, or ischemia-reperfusion injury [9]. In addition, changes caused characteristically by DM have been identified, such as alterations in the coagulation-fibrinolysis system in the form of hypercoagulability and vascular endothelial damage [6].

The most frequently affected zone by DMN is the anterior aspect of the thigh, followed by its posterior aspect [10]. The typical clinical presentation is acute-onset unilateral pain in the affected muscle, particularly the quadriceps femoris, local swelling, and subsequent induration of the tender area upon palpation. Previous trauma, fever, and sensory alterations are unusual, though some atypical presentations may be subacute and include motor deficiency with inability to stand and walk [3], and even bilateral presentations involving distal muscle groups [4]. The patient's clinical presentation was typical, manifesting sudden, severe unilateral pain and swelling in the left thigh in the absence of fever, along with typical physical examination findings.

The diagnosis of DMN consists first of establishing clinical suspicion of this pathology, keeping in mind that it is prevalent in patients with a history of poorly controlled diabetes, presenting with acute pain and unilateral inflammation of the proximal lower extremities not associated with traumatic events, causing weakness and limiting gait [3]. The two-month diagnostic delay observed in this case reflects a common pattern reported in the literature, attributable to the rarity of diabetic myonecrosis and its capacity to mimic infectious conditions such as pyomyositis or abscesses. As the patient's clinical course demonstrates, ultrasound can be nonspecific, leading to prolonged and ineffective antibiotic treatments. This finding emphasizes the need to consider early MRI in any diabetic patient with severe muscle pain unresponsive to initial antimicrobial therapy.

Likewise, DMN often presents with elevated serum biomarkers such as C-reactive protein and creatine kinase (CK) [7], although these findings are nonspecific, both of which were present in this case.

Soft tissue ultrasound often shows changes reflecting muscle inflammation, but these findings can be nonspecific and insufficient to establish a precise diagnosis [3]. Although ultrasound was the first imaging study requested for the patient, it only reported an inflammatory process without sufficient evidence to suspect DMN. In this context, MRI is the imaging modality of choice, showing perifascial, perimuscular, and subcutaneous edema coupled with hyperintense signals in the affected region on T2-weighted images and isointense or hypointense signals on T1-weighted images due to free interstitial fluid [3], which was shown in the imaging results.

If previous tests are inconclusive, a tissue biopsy can be performed, which has a 95% specificity showing edema, ischemia, and necrosis in early stages and fibrotic tissue with muscle fiber regeneration and lymphocytic infiltration in

more advanced stages [3]. However, it is usually contraindicated due to frequent complications such as poor wound healing, infection, seroma, and hematoma [4], leading to a longer recovery time [3], which is the reason why it was not performed in this patient's management.

In the differential diagnosis, deep vein thrombosis (DVT) is usually considered due to tenderness and lower limb swelling, but induration of the affected area points toward a muscular etiology in ischemic myositis [4]. Other considered diagnoses include abscesses, necrotizing fasciitis, muscle sarcomas, and pyomyositis; all of these can be excluded with a detailed history and imaging studies [3]. In the present case, the initial diagnostic suspicion was pyomyositis, which was ruled out following a lack of favorable clinical progress with antibiotic therapy and non-congruent MRI findings.

Treatment focuses on rest, analgesics, and strict glycemic control to prevent recurrences. NSAIDs and acetylsalicylic acid are effective in reducing recovery time; however, patients with this condition frequently suffer from diabetic nephropathy, limiting their use [3]. Their administration in this patient was strictly contraindicated due to the concurrence of a pharmacological allergy and stage 5 CKD, making it necessary to opt for alternative analgesic regimens.

The use of glucocorticoids in patients with poorly controlled type 1 Diabetes Mellitus represents a clinical challenge due to the risk of inducing severe hyperglycemia [11]. In this case, the prescription of deflazacort (6 mg/day) was justified as a short-term rescue anti-inflammatory measure against massive muscle edema and the impossibility of using NSAIDs due to renal failure and drug allergy. This intervention required close monitoring of the glycemic profile to ensure patient safety.

Although DMN usually resolves in the short term with proper treatment [3], the long-term prognosis is unfavorable. Recurrences are

estimated in 34.9% to 47.8% of cases months later, usually in the contralateral limb, or even exceeding 50% in the span of one year. Furthermore, the majority of DMN patients die within five years due to diabetes complications, including a high risk of infection and cardiovascular diseases [5], making close follow-up imperative [8]. In this case, the patient developed penile necrosis eight months later, a severe microvascular diabetic complication leading to infection, sepsis, and ultimately death.

Conclusions

In conclusion, diabetic myonecrosis is an infrequent complication of Diabetes Mellitus, in which diagnostic suspicion must be established in long-standing diabetic patients with suboptimal glycemic control, when unilateral pain and swelling in the limbs is manifested, predominantly in the proximal segments of the lower extremities. Magnetic resonance imaging stands out as the diagnostic method of choice due to its high sensitivity and specificity; its combination with anamnesis and the identification of microvascular risk factors such as retinopathy, nephropathy, or neuropathy facilitates diagnostic confirmation. Healthcare professionals must maintain a high index of suspicion for this unusual clinical entity and adopt a multidisciplinary approach to ensure timely and appropriate diagnosis and treatment.

Ethical considerations

The patient's personal information was anonymized and his relatives were informed about the nature and objectives of the research, with the study being conditioned on the voluntary acceptance of informed consent. Autonomy was respected as a bioethical principle in this case by allowing withdrawal from the process at any of its stages.

Conflict of interest

The authors declare that they have no financial, personal, or institutional conflicts of interest that could have influenced the results of this study.

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