



Contents lists available at ScienceDirect

Journal of Hand Surgery Global Online

journal homepage: www.JHSGO.org

Case Report

Spontaneous Diabetic Myonecrosis Presenting as Acute Carpal Tunnel Syndrome



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ARTICLE INFO

Article history:

Received for publication April 20, 2021
 Accepted in revised form October 20, 2021
 Available online 18 November 2021

Key words:

Carpal tunnel syndrome
 Hand
 Inflammation
 Muscle infarction
 Spontaneous diabetic myonecrosis

A 57-year-old man with diabetes mellitus presented with a 4-day history of left palm pain out of proportion, with swelling, erythema, and dense median and ulnar nerve distribution sensory changes. Magnetic resonance imaging with and without contrast revealed diffuse hand edema and myonecrosis. The patient was treated surgically because the examination was concerning for acute carpal tunnel syndrome and ulnar nerve compression. Spontaneous diabetic myonecrosis is a complication of diabetes mellitus that can be confused with several other conditions. It presents as acute-onset painful swelling in any muscle, and in the hand, may cause compressive neuropathies that necessitate surgical intervention.

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Spontaneous diabetic myonecrosis (SDM) is a rare complication of both type 1 and type 2 diabetes mellitus (DM) that involves the acute ischemic necrosis of skeletal muscle in the absence of a major arterial occlusion or atheroembolism. Diabetic myonecrosis is typically seen in patients with long-standing DM and may have other microvascular complications such as retinopathy, neuropathy, and nephropathy.^{1,2} It presents with an acute onset of painful swelling that progresses over days or weeks and can occur without any antecedent trauma or infection.³ Spontaneous diabetic myonecrosis is often mistaken for polymyositis or rhabdomyolysis. The most common location of SDM is the thigh, and it rarely presents in the upper extremity.⁴ We present a case of SDM in the hand with concomitant acute carpal tunnel syndrome requiring surgical intervention.

Case Report

A 57-year-old homeless man with a long-standing history of type 2 DM and multiple transient ischemic attacks presented to the emergency department with a 4-day history of severe pain and

swelling in his left hand. He described his symptoms as constant, throbbing pain, rated as a 10 out of 10 in severity, especially involving the thenar eminence and median nerve distribution of the hand, with slight radiation up his arm. His pain was spontaneous in onset and worsened over 4 days before presentation. Generalized wrist and digital movement, especially that of the thumb, worsened the pain. The patient denied any trauma or heavy exertion before the onset of his pain. He also complained of a progressive dense numbness on the palmar aspect of the hand in all 5 digits. He denied any preexisting neurological symptoms in the left hand, as well as any recent travel or febrile illnesses. He did not have any pain in any other muscle groups or symptoms of fevers, night sweats, or chills.

His past medical history included asthma, congestive heart failure, depression, DM with microvascular complications (eg, retinopathy), hypertension, obstructive sleep apnea, and transient ischemic attacks. The patient smoked approximately 5 cigarettes daily for more than 20 years. The drug screen was negative.

The patient was morbidly obese (body mass index of 42.3) and hypertensive with a blood pressure of 147/86 mm Hg. He was alert, oriented, and afebrile in the emergency department. His palm was diffusely erythematous and swollen but without fluctuance, and he was maximally tender over the thenar eminence. There was no tenderness along the flexor tendon sheath or pain on the passive stretch of the thumb or remaining digits. All compartments were soft and compressible, except for the thenar eminence, which was

Declaration of interests: No benefits in any form have been received or will be received related directly or indirectly to the subject of this article.

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<https://doi.org/10.1016/j.jhsg.2021.10.006>

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Figure 1. Clinical image of the patient at presentation showing a diffusely erythematous and edematous left hand.

firm but still compressible. There was no pain while performing short arcs within any joints of the hand and wrist. The 2-point sensation was absent at less than 10 mm in all the digits of the left hand compared with 5 mm throughout the contralateral hand. A Durkan's compression test was positive, suggesting carpal tunnel syndrome. The patient's pain precluded accurate manual muscle testing in the hand. A clinical picture of the hand is shown in [Figure 1](#).

Laboratory studies showed a normal white blood cell count of $9.33 \times 10^9/L$ with a normal differential, mildly low hemoglobin of 12.4 g/dL, and normal platelets of 242 K/uL (normal range, 150–399 K/uL). He had an elevated erythrocyte sedimentation rate of 25 mm/h (normal range, 0–17 mm/h) and C-reactive protein of 24.3 mg/L (normal range, 0–8.0 mg/L). His glucose was elevated to 151 mg/dL, but hemoglobin A1C was not collected. He had an elevated creatine kinase of 390.0 U/L (normal range, 10–205 U/L).

Radiographs showed only soft tissue swelling without soft tissue gas. Left upper extremity doppler ultrasonography did not display any evidence of deep vein thrombosis (DVT). A magnetic resonance imaging (MRI) scan of the left hand without contrast demonstrated heterogeneous T2 hyperintensity throughout the musculature of the hand, greatest in the thenar component ([Fig. 2](#)). There were areas of decreased T1 and T2 signal intensity centrally within the thenar musculature and to a lesser extent within the hypothenar musculature. There were no areas of abnormal T1 or T2 signals that would be consistent with osteomyelitis ([Fig. 3](#)). There was no associated hyperintensity of the skin overlying the thenar compartment. As per the radiologist's recommendation, a post-contrast study was obtained and revealed diffuse hyperenhancement indicative of edema and myositis throughout the musculature of the hand. There were areas of nonenhancement in the thenar musculature, and to a lesser extent, the hypothenar musculature, which is diagnostic of SDM. There was subcutaneous edema noted along the dorsum of the hand extending into the bases of the fingers distally. No rim-enhancing fluid collection was identified within the subcutaneous soft tissues that would suggest an abscess ([Fig. 4](#)).

Because of the progressive neurological decline, there was a concern for acute compression at the carpal tunnel and Guyon's canal. The patient underwent an emergent carpal tunnel release

and Guyon's canal decompression through a single volar incision. Edematous tissues were encountered intraoperatively, and the median and ulnar nerves appeared inflamed with a firm texture. Intraoperative cultures of the palmaris brevis muscle and deep carpal tunnel tenosynovium ultimately returned negative for microbiology and histopathology. Over the first 3 days, the patient noted a dramatic improvement in his hand pain and some improvement in his loss of sensation. The patient was discharged on postoperative day 3 after a steady improvement in his symptoms and normalization of laboratory markers. He was instructed to return to the clinic in 1 week; however, he was unfortunately lost to follow-up.

Discussion

Spontaneous diabetic myonecrosis, also known as diabetic muscle infarction, is a rare sequela of poorly controlled, chronic DM. Patients with SDM often have concomitant micro- or macrovascular complications of diabetes. The first description of SDM was in 1965 by Angervall and Stener, and at least 60 cases have been reported since then.⁵

While the pathophysiology of SDM is still unknown, it is believed to have a vasculopathic etiology, likely from the nonenzymatic glycosylation of endothelial proteins. Tissue ischemia from the damaged microvasculature triggers the inflammatory cascade, leading to ischemic necrosis. Subsequent reperfusion of necrotic tissues generates reactive oxygen species, inflammatory mediators such as tumor necrosis factor, and platelet-activating factor, which further promulgate vasculopathic changes. A hypercoagulable state may also be seen in diabetic patients because of alterations in the coagulation–fibrinolysis system, which may be a risk factor for this condition.⁴

The presentation of SDM is characterized by sudden pain and swelling of the affected muscle.³ While the thigh and lower extremity are most frequently affected, any muscle can be subjected. For example, Salehi et al⁶ reported a case of SDM in the neck musculature. We believe our patient constitutes the first report of SDM in the hand.¹ Patients will present afebrile and without systemic signs of infection. The affected muscle will be swollen and tender, and induration may be felt on physical examination.

There are commonly elevated but nonspecific laboratory findings, such as elevated plasma creatine kinase enzyme activity, erythrocyte sedimentation rate, and C-reactive protein.¹ Most patients with SDM have an elevation of any combination of these markers, and our patient had elevations in all 3.

Early noninvasive diagnosis of SDM is possible with MRI, and biopsy is rarely indicated.⁷ Specifically, a heterogeneously increased T2 or proton-density signal in an affected muscle, fascia, soft tissue parenchyma in a patient without clinical or laboratory features of infection is prototypical, which is consistent with our case history.⁸ Affected muscles will show modest gadolinium contrast enhancement.⁹

The differential diagnosis includes myositis (infectious or inflammatory etiology), DVT, or compartment syndrome. Myositis typically appears on MRI as smooth-walled intramuscular abscesses with rim-like enhancement and is less likely to present as acute-onset severe pain. While DVT and compartment syndrome do present as acute-onset severe pain, their history differs from SDM. Our patient did not have a history of trauma typical of compartment syndrome, nor intoxication and drug use typical of rhabdomyolysis. The clinical examination was also different, as there was no pain on passive stretching and the hand compartments were soft and compressible. Meanwhile, on MRI a DVT would have luminal filling defects with contrast, perivascular

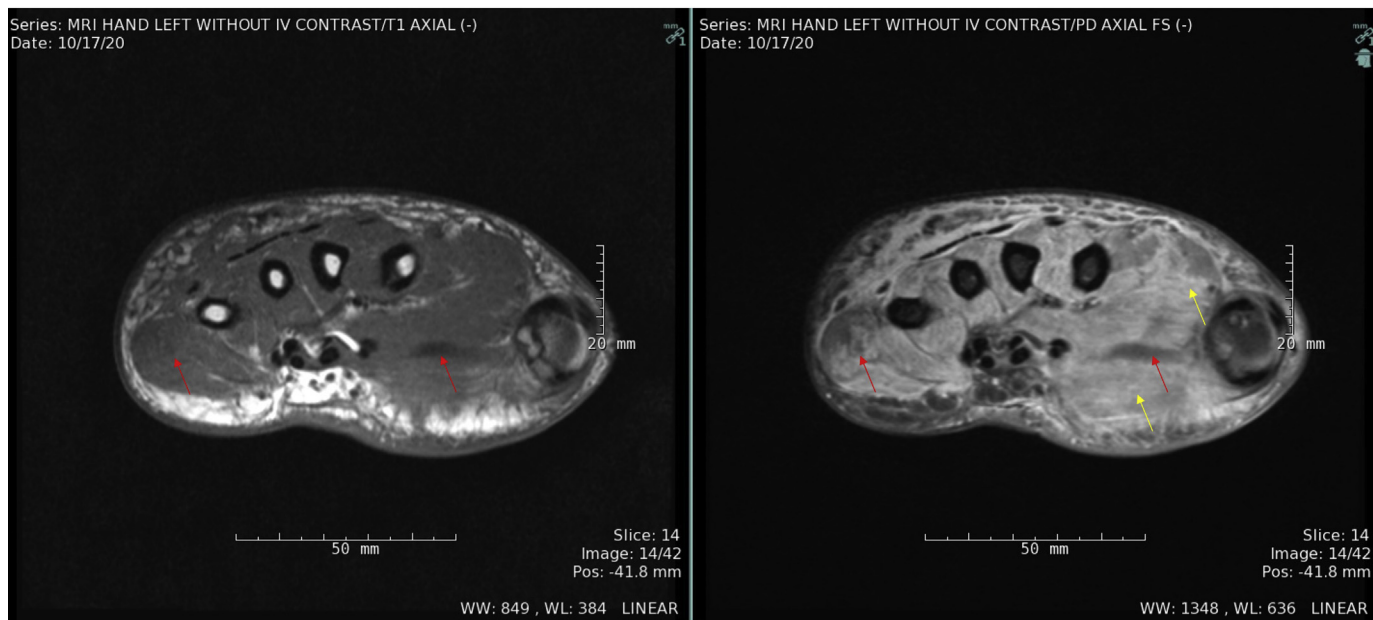


Figure 2. Heterogenous hyperintensity throughout the hand musculature on proton density fat-saturated MRI sequences suggest muscle edema (yellow arrows). Central areas of T1 sequence hypointensity in the flexor pollicis brevis and opponens digiti minimi muscles suggest infarction and myonecrosis (red arrows).

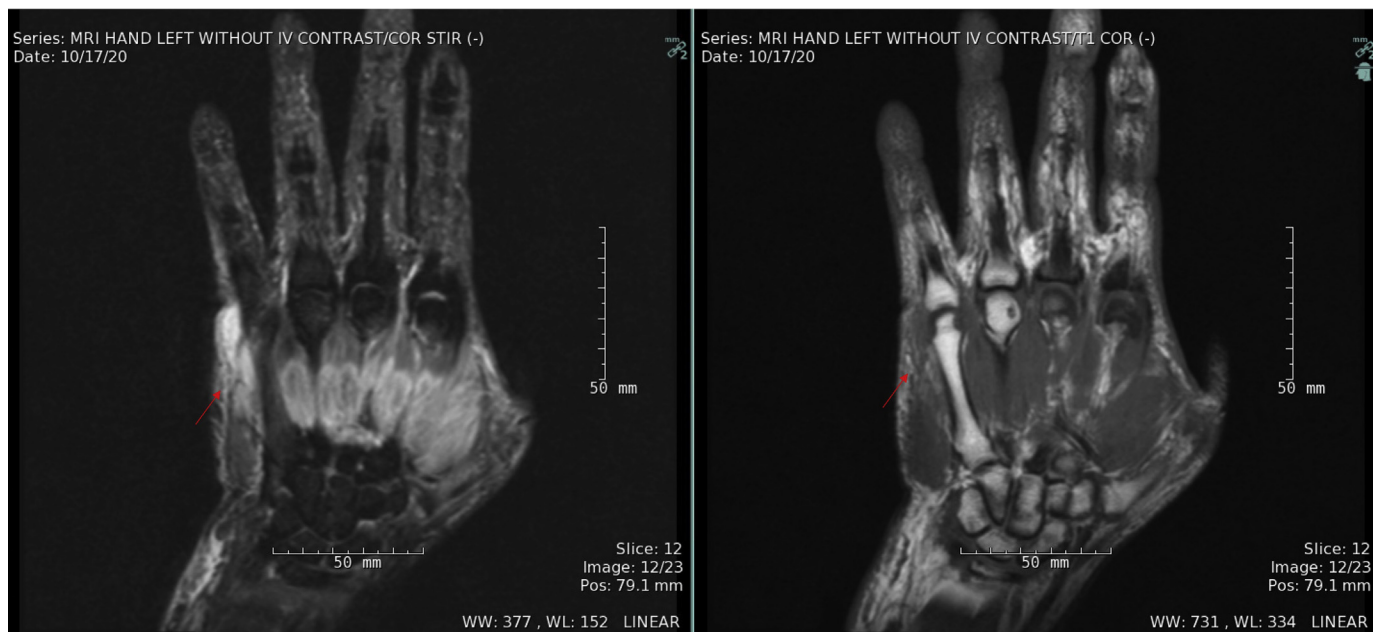


Figure 3. Subcutaneous soft tissue edema is present along the dorsum of the hand, extending into the bases of the fingers and along the ulnar side of the wrist (red arrows).

edema, and dermal thickening secondary to venous outflow obstruction.¹⁰ Our patient did not have these findings and was also negative on venous ultrasonography.

The treatment of isolated SDM is nonsurgical and relies on symptom, glycemic, and pain control. No optimal approach has been identified because of the limited number of reports of this condition. Also, because of the vasculopathic etiology of SDM, patients must be treated with an antiplatelet strategy involving low-dose aspirin or clopidogrel. Some patients may be eligible to receive nonsteroidal anti-inflammatory drugs (NSAIDs) for pain and symptom relief. Pain and symptom management for other patients not meeting these criteria should involve non-

NSAID agents, such as acetaminophen or low-dose opioid analgesics.

To our knowledge, this presents the first case of SDM reported in the hand, and it should be noted that it presented not only with several of the hallmark features of SDM seen elsewhere but also with concomitant acute carpal tunnel syndrome and ulnar neuropathy that necessitated surgical intervention. This presentation may have been because of the profound edema, swelling, and inflammation accompanying our patient's condition. Compressive neuropathies secondary to palmar hand-and-wrist swelling may not be seen to the same degree elsewhere in the body, such as the thigh, where risks of peripheral nerve compression are lower. We

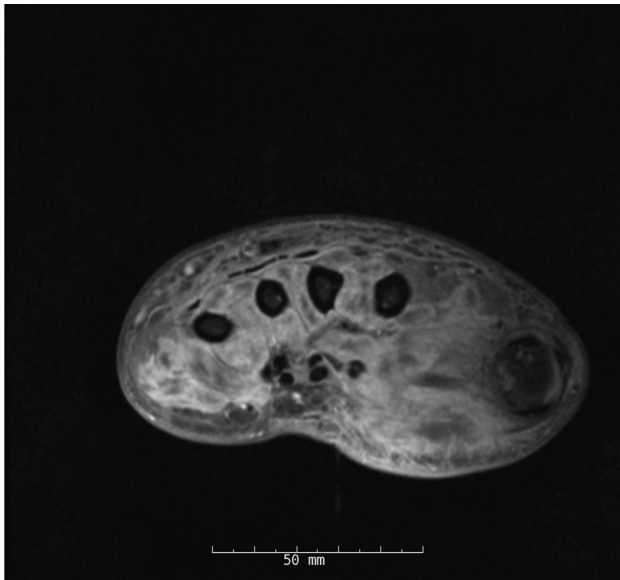


Figure 4. The addition of contrast fails to reveal ring-enhancing fluid collection, which would suggest myositis or an abscess in the musculature or subcutaneous soft tissues.

believe that it is essential for providers to have SDM on their differential diagnosis when working up atraumatic acute-onset pain and swelling in the hand in diabetic patients, and also must be aware of the risk of a resulting acute carpal tunnel syndrome that would require surgery. This clinical presentation may easily be confused with other inflammatory, infectious, or vascular conditions, each with different treatment strategies, thus making careful evaluation critical.

Unfortunately, our patient was lost to follow-up despite multiple efforts to reach him. Thus, we cannot determine if his neurologic symptoms fully recovered. However, postoperative findings in

the hospital showed rapid reductions in swelling, with the patient reporting considerable pain relief and some sensation return. While such follow-up information would have been useful, we believe it is valuable for clinicians to be aware of the combination of history, physical examination, laboratory studies, and imaging studies that created this unique presentation. Clinicians should also understand the appropriate treatment algorithm, including the management of evolving neurologic deficits.

Acknowledgments

The patient was informed and consented to allow photographs and case data to be submitted for publication.

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