

Single Case

# Painful Leg Ulceration in a Poorly Controlled Hypertensive Patient: A Case Report of Martorell Ulcer

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## Keywords

Martorell ulcer · Hypertensive ulcer

## Abstract

Martorell ulcer is a form of lower limb ulceration, preceded by a small area of excruciating pain. It often appears as a solitary lesion on the outer aspect of the lower limb, and is primarily associated with poorly controlled hypertension and diabetes. Treatment of the ulcer involves awareness and early correct diagnosis, adequate control of blood pressure, management of infection, and wound care. We describe a 77-year-old diabetic and hypertensive woman presenting with excruciating pain in her right lower lateral leg leading to a necrotic ulcer. Serial photographs of the evolution of the lesion and eventual healing of the ulcer are presented.

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## Introduction

The most common causes of lower limb ulcers include ischaemic ulcers, diabetic/neuropathic ulcers, and venous ulcers. Martorell ulcer is a rare ischaemic ulcer that is specifically associated with longstanding, poorly controlled hypertension [1]. It commonly affects women between the ages of 50–60 years. Typically, the patient presents with ex-

treme pain out of proportion to the size of the lesion. The pathophysiology is described as narrowing of small blood vessels in the skin which increases resistance to blood flow. The pathogenesis is associated with local factors triggering dermal arteriosclerosis and subsequent hyperplasia of the media layer and elastic lamina, a process known as hyalinosis [2]. This results in ischaemic changes leading to skin necrosis – hence ulceration [3–5]. The ulcer first appears as a solitary painful blister on the outer aspect of the lower leg [2] and eventually takes on an irregular deep appearance, which may lead to exposure of the underlying structures such as tendons.

### Case Report

A 77-year-old woman with a background of poorly controlled hypertension, type 2 diabetes mellitus, and peripheral vascular disease presented to the emergency department of a tertiary care hospital in May 2014 with an 11 × 7 cm necrotic ulcer on her right leg above the ankle. It was described as an irregular-shaped ulcer associated with areas of blistering and necrosis. The patient reported experiencing excruciating pain out of proportion to the size of the ulcer. The evolution of the ulcer and subsequent healing are depicted in the figures: evolving ischaemic ulcer before the breakdown of the epidermis at the outer region of the right lower limb (day 1) (Fig. 1); areas of necrosis and pus (day 5 after onset) (Fig. 2); patchy areas of soft tissue healing (day 14 after onset) (Fig. 3); well-healing ulcer after surgical debridement (day 18 after onset) (Fig. 4); formation of pink granulation tissue (day 44 after onset) (Fig. 5); after the split thickness graft (day 44 after onset) (Fig. 6); healed wound (October 2014) (Fig. 7).

The patient was hypertensive with systolic blood pressure ranging between 140 and 195 mm Hg and diastolic blood pressure between 65 and 97 mm Hg. Investigations revealed haemoglobin of 100 g/L, chronic renal impairment (creatinine 120 µmol/L, eGFR 38 mL/min/1.73 m<sup>2</sup>) and an elevated white cell count of 13.5 g/L (61% neutrophils). An autoimmune screen was positive for ANA. A wound swab was obtained which grew isolated colonies of *Staphylococcus aureus*. A subsequent bone scan showed increased vascularity of soft tissue in her right lower leg, without evidence of underlying osteomyelitis. In addition to this, a lower limb CT angiography and angiogram in the year preceding the ulcer showed bilateral 50% stenosis of the common iliac arteries and superficial femoral arteries. A lower limb angiogram performed after the ulcer showed high-grade stenosis in the right common and external iliac arteries requiring vascular stents to improve perfusion and healing.

This patient was managed with a multidisciplinary approach with input from specialties of vascular surgery, dermatology, microbiology, and cardiology. Medical management included analgesia and appropriate antihypertensives such as diltiazem and perindopril. Empirically, the patient received a course of intravenous Timentin to treat a mixed flora infection.

A radical debridement of the right lower limb ulcer was performed by the vascular surgical team. The necrotic tissue had full thickness to the level of the fascia and a clearly defined margin with adjacent healthy tissue. Tissue obtained during the procedure was sent for histopathological assessment which demonstrated ulceration with associated acute inflammation of the underlying dermis at the level of deep fascia. Additionally, vasculopathic changes were noted, including calcification of small vessels walls, intimal thickening, and recanalization with extensive acute inflammation. A vacuum-assisted closure dressing was applied. The patient also had known significant superficial femoral artery occlusive disease

and underwent a percutaneous femoral angioplasty to optimize perfusion 24 h after debridement. Aggressive inpatient and subsequent wound management was undertaken with the utilization of negative-pressure wound dressings. Once the ulcer bed had granulated sufficiently a split thickness graft was performed by the vascular surgeon. The skin graft was taken well and the ulcer healed completely.

## Discussion

In the case described above, the diagnosis of Martorell ulcer was made based on a combination of clinical findings including preceding excruciating pain, uncontrolled hypertension, type 2 diabetes mellitus, and histopathological analysis. The key feature supporting the diagnosis of Martorell ulcer in this case was based on the initial appearance of a small lesion on the right leg which was described to be extremely painful out of proportion to the size of the ulcer on a background of poorly controlled hypertension. The patient also had a history of diabetes mellitus, which has also been described in patients with hypertensive ulceration [6, 7]. The patient responded remarkably well to treatment of her high blood pressure with calcium channel blockers, which was the drug of choice in this scenario. Furthermore, histopathological features of the ulcer demonstrated calcification of small vessel walls in some areas, intimal thickening in other areas, and re-canalization, which strongly supported the diagnosis of Martorell ulcer.

Other differentials that were considered at the time of presentation were an ischaemic ulcer, calciphylaxis, and pyoderma gangrenosum. The clinical presentation of pyoderma gangrenosum is similar to that of Martorell ulcer. However, the features against a diagnosis of pyoderma gangrenosum are the absence of other predisposing associations, not having a typical purplish undermined edge, and gradual healing with treatment of hypertension. We feel that it is important not to misdiagnose this type of ulcer as pyoderma gangrenosum, because immunosuppressive treatment with prednisolone as for pyoderma gangrenosum would have aggravated her condition.

The patient was known to have peripheral vascular disease which was a concern for a developing ischaemic ulcer. However, in the context of an irregular-shaped ulcer with preceding excruciating pain, absence of peripheral ulceration in the feet, persistent hypertension, and histological features consistent with Martorell ulcer the diagnosis of a Martorell ulcer was made. The presence of risk factors for peripheral vascular disease, in particular uncontrolled hypertension, was certainly contributory to the formation of a Martorell ulcer. The patient had stage 3 chronic kidney disease secondary to chronic hypertension. Although kidney disease is also a feature in calciphylaxis, those patients are more likely to present with end-stage renal disease. Moreover, calciphylaxis typically presents with purple-coloured mottling of the skin with the occasional appearance of blood-filled blisters, which were absent in this case [8]. In addition, the histopathology did not support a diagnosis of calciphylaxis in this case.

Although atypical infections were also considered in the differential diagnosis, microbiological investigations, histopathology, and the evolution of the ulcer did not support this possibility. This patient had a very successful outcome with complete wound healing subsequent to appropriate wound care and timely skin grafting. It is important to make the correct diagnosis in Martorell ulcers for optimal management, particularly as the main differential diagnosis is pyoderma gangrenosum, which has a completely different approach in treatment.

## Conclusion

This case emphasizes the importance of considering Martorell ulcer in patients presenting with a lower leg ulcer on a background of poorly controlled hypertension and disproportionate excruciating pain in the ischaemic area prior to ulceration.

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## Statement of Ethics

This case report satisfied local ethics guidelines and informed consent was obtained from the patient.

## Disclosure Statement

The authors have no conflicts of interest to declare.

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**Fig. 1.** Evolving ischaemic ulcer before the breakdown of the epidermis at the outer region of the right lower limb (day 1).



**Fig. 2.** Areas of necrosis and pus (day 5 after onset).



**Fig. 3.** Patchy areas of soft tissue healing (day 14 after onset).



**Fig. 4.** Well-healing ulcer after surgical debridement (day 18 after onset).



**Fig. 5.** Formation of pink granulation tissue (day 44 after onset).



**Fig. 6.** After the split thickness graft (day 50 after onset).



**Fig. 7.** Healed wound (October 2014).