



Painful leg ulceration: A case report and concise review of symmetrical Martorell's ulcers

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Abstract

Martorell's ulcer is a rare ischemic and extremely painful lesion located in the distal portion of the lower limb. It is common in women and resulting from severe systemic and poorly controlled hypertension. Treatment of the ulcer involves awareness and early correct diagnosis, adequate control of blood pressure, management of infection, and wound care. Case presentation: A 58-year-old female Brazilian patient presented with an ulcer on the right lateral calf, which was extremely painful. For the treatment of Martorell's ulcer, we used the combination therapy, which included antihypertensive and antiagregant drugs. After that was noted intense regenerative processes and recovery of the patient. Conclusion: Martorell's ulcer requires the complex and aggressive approach to the treatment under obligatory antihypertensive therapy and should always be considered in the differential diagnosis of painful lesions of the lower limbs especially in women.

Keywords

Martorell ulcer, Hypertensive ulcer, chronic leg ulcers, atypical ulcer

Introduction

First described by Martorell and supported by Hines and Farber from the Mayo Clinic, hypertensive leg ulcer or Martorell ulcer is defined by progressive and painful necrotic ulceration, pain disproportionate to the size of the ulcer, specific location on the lower two-thirds of the leg, absence of significant arterial or venous disease and associated with long-standing, poorly controlled hypertension [1, 2]. It commonly affects women between the ages of 50–60 years [3-5]. Approximately 50% of patients have bilateral involvement, sometimes occurring months to year's apart [6].

Typically, the patient presents with disproportionate pain out of proportion to the size of the lesion [1-3]. The pathophysiology is described as the narrowing of small blood vessels in the skin which increases resistance to blood flow [2, 3]. The pathogenesis is associated with local factors triggering dermal arteriosclerosis and subsequent hyperplasia and obstruction of the small arterioles of the medial artery [3].

The clinical criteria described by Martorell for the diagnosis of hypertensive ulcer were: location on the inner side of the distal third of the lower limbs; diastolic arterial hypertension in the lower limbs; 'hyper pulsatility' of the arteries of the lower limbs, absence of arterial calcifications; absence of chronic venous insufficiency; symmetrical lesions or residual hyper pigmentation of previous ulceration in the inner side of the inferior limb; increased pain in the horizontal position, with greater prevalence in women [3, 6, 7].

We present a case of Martorell's hypertensive ischaemic leg ulcer in a woman of long-standing and poorly controlled essential hypertension who presented a large ulcer on the right calf and a concise review of the literature.

Case Presentation

A 58-year-old woman presented with complaints of long-standing, non-healing, and painful ulcers on the left leg and one small, recently healed ulcer on her right leg. Before her first visit to the authors' clinic, the patient had been given a diagnosis of pyoderma-gangrenosum. The patient had a 30-year history of chronic hypertension

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with blood pressure values up to 210/120 mmHg and she had been receiving treatment without effect for 4 months at a different clinic. History disease revealed blood pressure elevations since 35 years old age, but the patient sought medical advice extremely seldom and did not take antihypertensive treatment. She was examined for renal dysfunction, endocrine disorders, and vascular disease as the potential cause of hypertension, but without any pathologic findings. She noted when was 52 years old a painful ulcer was located on antero-lateral surface of the right leg but now it's healed (Figure 1). The condition deteriorated in 2019 when the ulcer defects become larger and more painful on her left leg.

On admission at the examination, the blood pressure was 210/100 mmHg in the left arm sitting and 190/95 mmHg in the right arm sitting. The ulcer of an oval shape (size 8.0 × 3.0cm and depth 0.3 cm) on the lower third of the calf of the left leg is present (Figure 2). The base was covered by fibrinous exudate with some buds of granulation and sharply demarcated margins. The skin around the ulcer was hyperemic and swollen with local hyperthermia. There were no signs of venous stasis. The posterior tibial artery and dorsalispedis artery pulse were palpable.

In clinical and biochemistry analyses no pathology was found. X-ray of the right foot and ankle was negative for osteomyelitis. Arterial occlusive disease of the lower limbs was excluded by color Doppler duplex examination. WBC 8000, haemoglobin 12.8 g/dL, fasting glucose 87 mg/dL, serumcreatinine 1.1 mg/dL and homocysteine level was normal. There were signs of left ventricle hypertrophy on the data of electro and echocardiographic investigations.

With the purpose of diagnosis specification, pathomorphologic study of tissue fragments of the injured zone was performed. The results of this study showed that revealed changes are relevant for hypertensive ulcers of legs or Martorell's ulcers.

At the beginning of treatment, the patient self-reported her pain using the visual analog scale (VAS; 1 = no pain, 10 = extremely painful) as a 9. After being prescribed opioid and antihypertensive her VAS reached 3.

Her hypertension was treated with antihypertensive medication (furosemide and valsartan), and her diastolic blood pressure stabilized to 80 mm Hg. After 4 weeks, the blood pressure became normal (120-140/90-80 mm Hg) and the size of her Martorell's ulcer reduced. The lesion became cleaner and partly epithelized and the VAS score reached 1.



Figure 1: The condition deteriorated in 2019, when ulcer defect become larger and more painful on her left leg.



Figure 2: The base was covered by fibrinous exudate with some buds of granulation and sharply demarcated margins.

The woman was discharged after improvement of the condition and she continued to take antihypertensive medication. Control examinations after 6 and 9 months showed no skin defects and slight soft scars. The patient was instructed to continue treatment with antihypertensive medication and to have periodic visits with the surgeon and cardiologist.

Discussion

The history of the study of hypertensive ulcers of the lower extremities begins in 1945 with the famous Spanish angiologist Fernando Otset Martorell who described a rare disease associated with chronic progressive hypertension which complicates ulcer defects on the extremities [1, 2, 8]. According to the literature, about 0.5% of the adult population in western countries still suffers from this pathology. This nosology occurs in no more than 5% of the total lower extremity ulcer and is reported to have an impact on virtually every aspect of daily life [8, 9].

Severe systemic arterial hypertension is the main etiologic factor provoking lower extremity ulceration related to Martorell ulcer ("a disease caused by another disease") [8]. Chronic wounds of the lower extremities can be of infectious, hematologic, or malignant etiology, but are most commonly caused by peripheral arterial occlusive disease [6].

These lesions are most commonly on the lateral aspects of the lower extremities and often symmetrical [5-7]. These ulcers usually have been present for long periods of time and found to be refractory to the usual local measures for ulcer healing [7].

This patient presented with the main clinical characteristics of Martorell's ulcer, such as chronic hypertension, the absence of clinically significant arteriosclerotic large artery occlusive or venous disease, and the presence of satellite lesions [1, 5]. In addition, several other criteria such as (*) the location of ulcer on the antero-lateral part of the lower leg, (*) the lack of chronic venous insufficiency, and (*) strong, increasing pain levels were also described by this patient [5-8].

The main pathogenetic factor in Martorell ulcer is an angiospasm and a stenosis of hemo-microcirculatory vessels [8].

There is a concentric growth of connective tissue in all layers of the wall with obliteration of their lumen causing endothelial proliferation and sub-endothelial-hyalinosis in hypertrophied arterioles [8, 9].

Venous leg ulcers are the most common cause of leg ulcers, accounting for 60-80% of them [3]. Normally located at the lower third of the leg (gaiterregion) and pain may be absent or is more at the end of the day and usually relieved by elevation of the leg. Painful lower extremity ulcers may occur in a variety of conditions such as differential diagnosis like vasculitis, calciphylaxis, pyodermmagangrenosum and skin thrombosis (thrombophilia, anti-phospholipidsyndrome, cryoglobulinaemia) [10]. The pain seen with hypertensive ischaemic ulcers is frequently reported to be far more severe than would be expected for the size of the ulcer and may even be the sole reason the patient seeks medical care [7, 10-12].

Our case is almost identical to Martorell's index case of hypertensive ischaemic leg ulcer, in particular typical location, presence of intense pain, the absence of arterial or venous pathology, and representative of the demographics of age and gender in the cases in the literature.

The usual methods of treating Martorell's ulcer are surgical treatments that involve debridement, skin grafting, and lumbar sympathectomy. All options associated with antihypertensive medication which help to reduce pathogenesis of ulceration development but the results of these treatments are variable [5, 8].

Wirsing *et al* in 2019 [5] reported an effective noninvasive approach to the treatment of Martorell's ulcer and Duzhiy *et al*. 2017 [8] described a complex therapy improved by the platelet-rich auto plasma activates the therapeutic effect, which results in intensive healing of the ulcer and faster improvement in pain relief.

Total ulcer healing requires the combination of various therapies. Reduction of blood pressure levels is critical to the success of the treatment and not only prevents increased vascular damage but improves blood supply through local vasodilation. The drugs of choice are calcium channel blockers and angiotensin-converting enzyme inhibitors [4].

Surgical treatments involve the debridement of devitalized tissue with second intention closure or skin grafting and lumbar sympathectomy. The result is variable, from complete recovery to little impact on healing [4, 6, 8].

Such ulcers usually present for long periods and are refractory to the usual topical therapies for ulcer healing. Although it is important to lower the patient's blood pressure to reduce further arteriolosclerosis, limited benefit can be derived from successful blood pressure control [5, 6, 8-10]. Multi-professional follow-up is very important and include cardiologist assistance.

In summary, Martorell's ulcers often an unrecognized cause of chronic leg ulcer, requires the complex approach to the treatment focused on optimizing antihypertensive therapy and should always be considered in the differential diagnosis of painful lesions of the lower limbs, especially in woman.

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