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Ultero-osteolytic lesions in a woman with type 2 diabetes and carpal tunnel syndrome: A case report and literature review

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ABSTRACT

We describe the case of a 73-year-old woman with type 2 diabetes presenting with ulcers and osteolytic lesions on distal phalanges of left hand, who was diagnosed with the rare “ultero-mutilating” variant of carpal tunnel syndrome. A review of literature on cutaneous manifestations associated with the syndrome is also presented.

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1. Case report

We describe the case of a 73-year-old woman with type 2 diabetes who presented with ulcerations and necrotic lesions involving the distal phalanges in the second and third fingers of the left hand.

The patient had been diagnosed with diabetes 10 years before; she was on metformin 2 g/day with an overall acceptable glucose control in the past.

She had a clinical history of total thyroidectomy for a nodular goiter, and she was on levothyroxine 125 mcg/day since then. She had hypertension and dyslipidemia, and is currently under treatment with irbesartan 150 mg/day and simvastatin 20 mg/day.

Cutaneous manifestations appeared roughly 12 months before and they were treated with antibiotics without any

clinical benefit. They first presented as painless bullous eruptions on the left hand located in the second and third phalanges, progressively evolving towards ulcerative and necrotic lesions and the mutilation of the second distal phalanx (Figs. 1–3). On the right hand, initial nail plate abnormalities of the second and third fingers were also evident (Fig. 4).

On both hands, thenar atrophy and severe hypoesthesia were detected, whereas Raynaud's phenomenon was absent. Palpation of the peripheral pulses revealed good radial and brachial pulses. On general physical examination, no other significant signs were observed.

At laboratory tests, complete blood count, coagulation parameters, renal function, protein profile and complete urine tests as well as C-reactive protein were all within normal range, except for a mild increase in erythrocyte sedimentation rate, ESR (23 mm/h). HbA1c levels (8.1%, 65 mmol/mol) revealed an inadequate glucose control.

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Fig. 1 – Lesions on distal phalanges of left hand.



Fig. 3 – Lesions on distal phalanges of left hand.

Screening for autoimmunity (anti-nuclear antibodies, anti-neutrophil cytoplasmic antibodies, anti-parietal cell antibodies, anti-Scl70 antibodies, anti-nDNA antibodies, anti-centromere antibodies, complement components C3 and C4) and neoplastic markers (CEA, CA 19-9, CA 125, CA 15-3 and α -fetoprotein) were negative.

She underwent a specialist rheumatologic, dermatological, plastic surgery counselling to exclude systemic diseases or cutaneous neoplasm.

A complete cardiovascular screening was also performed, including electrocardiogram, echocardiography and vascular Doppler ultrasound, which revealed the presence of atherosclerotic plaques, not determining hemodynamic alterations at carotids and lower limbs arteries. Absence of plaques and the presence of an adequate arterial vascular flow were detected at upper limbs arteries, also in the distal vessels of both hands.

Periungueal capillaroscopy did not indicate any pathological nail fold capillary change.

Hands radiographs showed bone resorption and osteolysis in the distal phalanx of the left index finger with loss of adjacent skin soft tissue and early bone resorption in the distal phalanx of the third finger (Fig. 5).



Fig. 2 – Lesions on distal phalanges of left hand.

The patient then underwent a complete neurological examination including electromyography which revealed a severe bilateral compression of median nerve and complete absence of motor and sensory conduction response to nerve stimulation.

Bilateral CTS diagnosis was therefore confirmed and the associated neurotrophic lesions suggested the rare necrotic variant of the syndrome.

The patient was then referred for bilateral surgical decompression of median nerve, that resulted in a complete resolution of cutaneous ulcero-necrotic lesions on left hand after 1 month of follow-up (Figs. 6 and 7).

2. Discussion and literature review

Carpal tunnel syndrome (CTS) is the most common entrapment neuropathy, and it usually presents with a classic triad of symptoms, including nocturnal pain, hypoesthesia and thenar atrophy [1].

In some cases, clinical presentation may be atypical and, in the late stages, a variety of skin manifestations such as erythema, oedema, blistering, sclerodactyly, cutaneous



Fig. 4 – Initial nails alterations on second and third fingers of right hand.

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