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Title page

Manuscript title: Digital ulcers as the first manifestation of carpal tunnel syndrome

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Abstract

While carpal tunnel syndrome is the most common entrapment neuropathy presented at rheumatologic consultation, its ulcerative variant is rarely described. We report of a case of severe bilateral medial nerve palsy, presented to us for diagnosis of chronic digital ulcers which had been suspected to be rheumatologic in origin. Careful examination revealed an unusual case of carpal tunnel syndrome in an advanced stage, which presented itself with skin changes only, lacking all the typical neurologic symptoms or rheumatologic background. Our case stresses the importance for rheumatologists to be aware of the ulcerative form of CTS as one of the etiologic differential diagnostic options of digital ulcers.

Key words: carpal tunnel syndrome, digital ulcers, acroosteolysis

Digital ulcers as the first manifestation of an advanced stage of carpal tunnel syndrome

K. Perdan-Pirkmajer, S. Praprotnik, M. Tomšič

Introduction

Carpal tunnel syndrome (CTS) is the most common entrapment neuropathy presented at rheumatologic consultation [1]. While most CTS are considered to be idiopathic or related to repetitive flexor wrist movements, the presentation of an evolving rheumatologic disease as a CTS is also very well known [1, 2]. The ulcerative and mutilating clinical variant of CTS was described for the first time by Bouvier and coworkers [3]. It occurs in patients with severe damage of motor, sensory and above all autonomic nerve fibers and usually presents in the advanced stages of disease [4, 5]. It is characterised by appearance of ulcerative skin lesions, skin atrophy and acroosteolysis in the sensory zones of the median nerve [5, 6] as well as nail changes such as onychodystrophy [7].

Case report

A 85-year old woman was referred to our Rheumatology department for diagnosis of chronic skin ulcers and blisters which had been reappearing spontaneously for the last 6 years on index and middle fingers bilaterally. She also noticed vesicular lesions beneath the nails, but apart from that, she had no other complaints. She has been treated for hypertension and osteoporosis with ACE inhibitors and biphosphonates for years. Her history of smoking and drinking was negative. She has worked on a farm most of her life.

Index and middle fingers were affected by ulcerations, which were not painful upon palpation (Fig.1). The same fingers were also affected by almost complete loss of the sense of touch. Nails on both index fingers were markedly dystrophic. There was a prominent bilateral thenar atrophy without any appreciable loss of strength in hand grip. Radial pulses were symmetrically palpable. Rheumatologic status was completely normal apart from signs of osteoarthritis in hands and feet. Laboratory tests revealed slightly elevated erythrocyte sedimentation rate, 24 mm/h (<15) and normocytic anaemia, haemoglobin 93 g/L (120-160), blood sugar level was normal, Hep-2, ENA, ANCA, cryoglobulines, aPL antibodies were all negative. Hands X-ray showed typical signs of osteoarthritis of small joints in both hands and bilateral acroosteolysis of distal phalanx of middle fingers. Median nerves were ultrasonically thinned and electrophysiologic findings were consistent with severe bilateral

median nerve sensori-motor neuropathy. Capillaroscopy revealed no abnormalities. During her stay at our department, most of her ulcers began to heal without any additional therapy (Fig.1).

Discussion

Our patient had typical neurologic deficits, sensori-motor neuropathy with thenar atrophy, skin lesions and onychodystrophy, X-ray evidence of acro-osteolysis and lack of any clinical or laboratory data suggesting inflammatory rheumatologic disease. This was all consistent with severe CTS. Her occupational history also suggested the possibility of an entrapment neuropathy due to repetitive wrist movements, while normal blood sugar level and radial pulses spoke against diabetes or ischemia contributing to the clinical picture.

Although skin involvement may present in up to 20% of CTS, there have been but a few reports on it, mostly in dermatologic and surgical literature [4, 7]. Nevertheless, this is, to our knowledge, the first reported case of a patient with ulcerative variant of CTS, who presented with digital ulcers in an advanced stage of disease, while neurologic signs, which must have developed much earlier, went completely unnoticed by the patient. All the typical symptoms like unremitting pins and needles, dropping objects and nocturnal pain were absent. Apart from that, ulcerative variant of CTS occurs more often in men and is bilateral in only 25% of cases [8].

Our case stresses the importance for rheumatologists to be aware of the ulcerative form of CTS as one of the etiologic differential diagnostic options of digital ulcers.

Disclosures: The authors declare no conflict of interests.

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Figure caption:

Fig1 Digital ulcers on index and middle fingers before (upper insert) and after (lower insert) hospitalisation.

