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Case Report

Elephantiasis nostras verrucosa associated with psoriasis in a Chinese patient

疣狀本土性象皮病伴發銀屑病：華人病例一宗

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A 52-year-old Chinese man with twenty years of psoriasis presented with characteristic cobblestone-like lesions over his legs. Biopsy of the lesions showed pseudo-epitheliomatous hyperplasia typical of elephantiasis nostras verrucosa. This is the first reported case of elephantiasis nostras verrucosa associated with psoriasis in a Chinese.

52歲男性華人患者患銀屑病有20年，雙腿出現特徵性的鵝卵石狀皮損。皮膚活檢診斷為假上皮瘤狀增生，此為疣狀本土性象皮病的典型病理組織學變化。本例為首宗華人疣狀本土性象皮病伴發銀屑病病例報告。

Keywords: Chinese, elephantiasis nostras verrucosa, psoriasis

關鍵詞：華人，疣狀本土性象皮病，銀屑病

Introduction

Elephantiasis nostras verrucosa (ENV) is a rare chronic disorder with characteristic cutaneous papillomatosis and hyperkeratosis, and manifesting as pebbly, verrucous and cobblestone-like lesions over thickened skin. The histological change is pseudo-epitheliomatous hyperplasia. The aetiology of ENV is thought to be related to chronic lymphatic insufficiency. The diagnosis is based on typical morphological changes and histological finding of pseudo-epitheliomatous hyperplasia. Two cases of ENV associated with psoriasis have been reported in Scotland and Japan respectively.[1,2] The following is a case report of a 52-year-old Chinese male patient with ENV and psoriasis.

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Case report

A 52-year-old Chinese man with psoriasis vulgaris attended dermatological clinic for twenty years,

but frequently defaulted follow-up. He also gave a history of recurrent skin infection accompanied with progressive but gradual swelling of both of his lower legs for a couple of years. He was admitted into hospital for his extensive plaque psoriasis and methotrexate was given at 15 mg orally per week. The psoriatic plaques responded well and most psoriatic plaques resolved in four weeks.

However, multiple non-scaling, skin-coloured papules and nodules remained, covering the non-pitting swollen legs from feet dorsa to the upper shins (Figure 1). The individual papillomatous lesions were fleshy, verrucous and of different sizes, confluencing to form cobblestone-like, firm, verrucous thickened plaques (Figure 2). As the lesions differed considerably in morphology from that of psoriasis, an incisional skin biopsy was

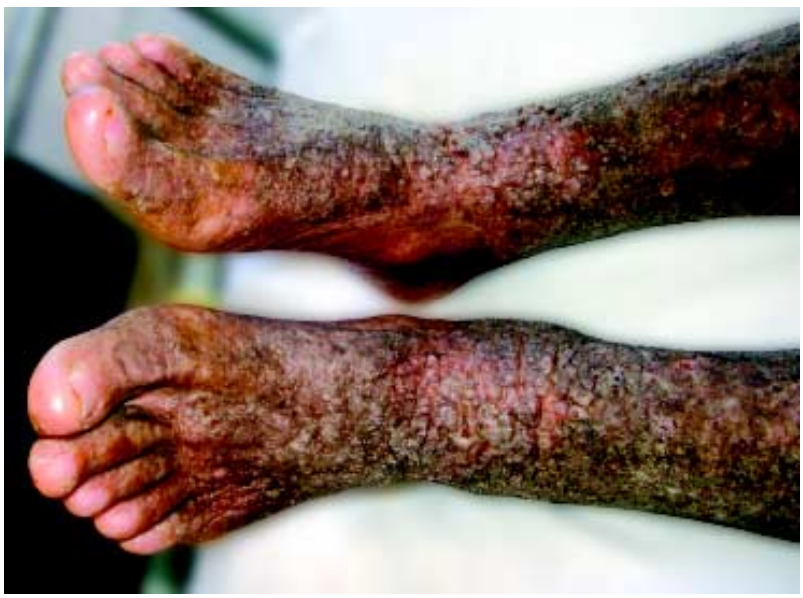

Figure 1. Verrucous plaques covering the anterolateral aspects of both lower shins and feet dorsa.

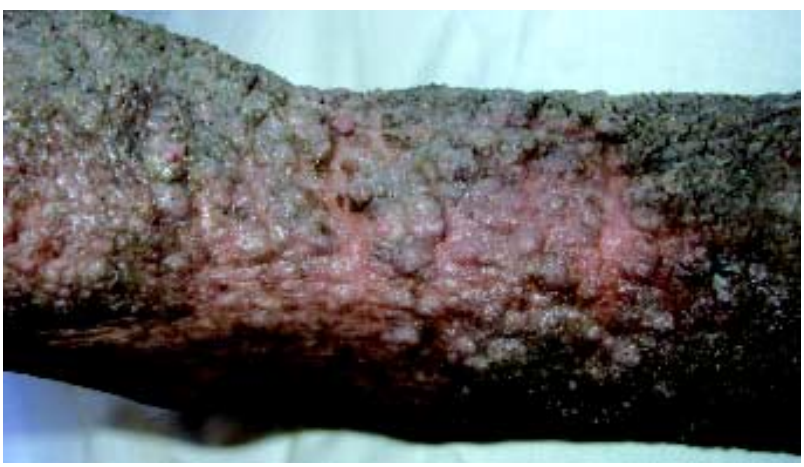

Figure 2. Close-up view of right ankle showing confluencing cobblestone-like papulonodules.

performed. The histology showed pseudo-epitheliomatous hyperplasia (Figures 3, 4 & 5). Given the past history of recurrent infection and swelling of his lower legs, the characteristic cobblestone-like lesions and histological findings, the diagnosis was compatible with ENV.

Discussion

Chronic peripheral oedema due to venous or lymphatic stasis of diverse origin may produce thickened skin with hyperkeratotic verrucous and papillomatous lesions; these changes are known as elephantiasis.[3,4] Traditionally, "elephantiasis" has been used to describe the cutaneous changes that result from chronic filarial lymphatic obstruction caused by Wuchereria bancrofti, W. malayi or W. pacifica infestation.[5] In 1934, Castellani introduced the adjective "Nostras", which means "ours", "of ours", "of our region" (temperate zone), differentiating this condition from the filarial elephantiasis tropica. [3]

Price[6] described two types of ENV. One is "soft" or "water-bag" type and the other "hard" or "wooden" type. In the "water-bag" type, there is smooth and thickened skin, which is freely movable; whereas in the "wooden" type, there is less swelling, and the fibrotic skin is fixed to the deeper structures, and, in some of them, with development of multiple verrucous firm nodules, as seen in our patient.

The common mechanism for elephantiasis is thought to be due to underlying chronic lymphatic stasis, which can occur under numerous conditions, including bacterial infection, trivial trauma, tinea pedis, Streptococcal and Staphylococcal infections, congenital or surgical lymphatic disruption, venous stasis, radiation, neoplastic obstruction, portal hypertension, obesity and congestive heart failure.[7-10] It was thought that psoriatic lesions could cause local lymphatic

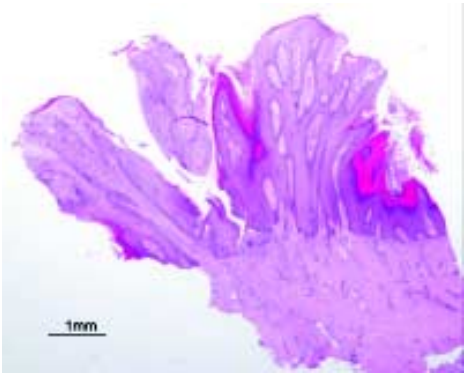

Figure 3. Low power view showing marked pseudoepitheliomatous hyperplasia. (H&E x 10 original magnification)

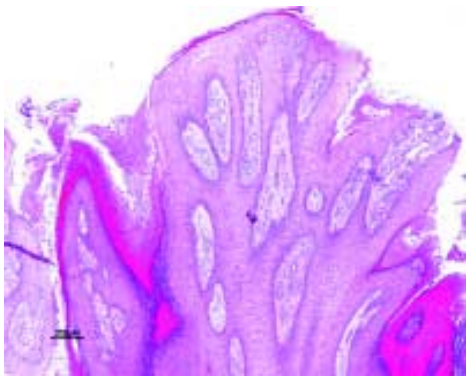

Figure 4. Note the striking hyperkeratosis and acanthosis. Mild inflammatory cell infiltration is also noted in the papillary dermis. (H&E x 40 original magnification)

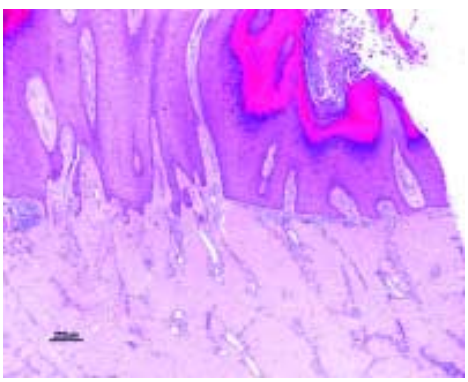

Figure 5. There is fibrous scarring in the dermis with horizontally arranged collagen and vertically arranged blood vessels. (H&E x 40 original magnification)

disturbances leading to development of stasis papillomatosis.[2]

The typical histological finding of ENV is pseudo-epitheliomatous hyperplasia.[4,9] Papillomatosis cutis carcinoides of Gottron, a condition mimicking ENV, shared the common histological findings of pseudo-epitheliomatous hyperplasia. While ENV runs a benign course, papillomatosis cutis carcinoides may undergo malignant transformation.

The diagnosis of ENV is based on the history and clinical examination.[5,10] A history of recurrent attacks of lymphangitis, trauma, surgery or radiotherapy may be found in many cases. Histopathology and lymphatic imaging may provide further information when the clinical features are not diagnostic.[5,10]

The treatment of ENV is difficult and aims at restoring the function of the limbs, rendering the patient socially acceptable and reducing incidence of infection, minimising the chance of amputation. The underlying disease causing ENV, such as congestive heart failure, liver disease, renal disease and hypoalbuminaemia should be treated. General measures include improvement of personal hygiene and nourishment, elevation of the affected limbs, and usage of pressure hosiery or pneumatic pump. Prompt treatment of superimposed infection is important. Systemic retinoids are useful in the treatment of diseases that are characterised by abnormal epidermal proliferation and lichenification. Impressive therapeutic effect of oral etretinate on ENV has been reported, with the dose of 0.6-0.75 mg/kg daily for four to six weeks.[1,4]

So far, there has been two reported cases of ENV associated with psoriasis found in the English literature.[1,2] To the best of our knowledge, this is the first reported case in a Chinese.

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