

中文題目：以疣狀非血絲蟲性象皮病表現之雙下肢深部靜脈血栓：個案報告

英文題目：Bilateral Lower Extremities Deep Vein Thrombosis Presented with Elephantiasis

Nostras Verrucosa : a case report

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Introduction:

Elephantiasis nostras verrucosa (ENV) is a rare complication of chronic lymphedema. We herein presented a case of an 89-year-old bedridden woman with ENV of bilateral lower extremities, which was caused by deep vein thrombosis and chronic venous insufficiency.

Case Presentation:

This is an 89-year-old woman with a history of type 2 diabetes mellitus and hypertension. Due to bilateral legs weakness, she became immobilized after receiving open reduction and internal fixation for left ankle malleolar fracture 5 years ago. 8 months prior to this admission, she claimed bilateral lower legs fullness and swelling with erythematous skin rashes, skin eruption and numerous verrucous protruding masses. She had been brought to several local clinics, and been treated with oral antihistamines and topical corticosteroid ointments. The symptoms did not improve, so she was sent to our hospital for further evaluation.

On admission, her temperature was 36.6°C and vital signs were stable. Physical examination revealed verrucose plaques and nodules with massive non-pitting edema over bilateral legs. Local heat was noted over her left lower leg (Figure 1). The Kaposi-Stemmer sign was not significant due to disfiguration of skin fold over the dorsum of her feet. Laboratory evaluation revealed white blood cell count of 12500/uL with elevated C-reactive protein level 10.06 mg/dL (normal, 0 to 5.0). Other abnormal results included elevated blood urea nitrogen of 31.0 mg/dL and D-Dimer of 1.82 mg/L FEU (normal, <0.55 mg/L FEU). Thyroid function tests, B-type natriuretic peptide and albumin levels were within normal limits. Urinalysis revealed no proteinuria by dipstick. Based on history and physical examination, diagnosis of ENV was made by dermatologist, and antibiotics administration was suggested as secondary infection is considered. Computed tomography angiography revealed thrombi in the right distal femoral vein, left popliteal vein and posterior tibial vein, associated with soft tissue swelling and fasciitis over bilateral lower extremities (Figure 2). Under the impression of deep vein thrombosis of bilateral legs, complicated with ENV and secondary cellulitis, treatments included intravenous amoxicillin-clavulanate and oral Rivaroxaban 15 milligram twice daily were initiated. Elevation of the affected limbs while lying on the bed, daily wound care of skin eruption, and massage were encouraged. After one week of treatment, erythema, swelling and lower leg pain had improved significantly (Figure 3).

Discussion:

ENV is a rare but severe complication of chronic nonfilarial lymphedema. It is characterized by non-pitting edema and superimposed hyperkeratotic papulonodules with a verrucose or cobblestone-like appearance.¹ Lower extremities are the most common site of ENV, but any area with chronic lymphedema can be affected such as the upper extremities, abdomen, and scrotum.²

The exact pathogenesis of ENV is still not clear, but a vicious cycle of chronic lymphedema and repeated infections is a very important contributing factor.³ Conditions including congenital or secondary to filarial, staphylococcal, and streptococcal infection, previous surgery or trauma, radiation, neoplastic obstruction, obesity, chronic venous insufficiency, portal hypertension, congestive heart failure, hypothyroidism play an essential role.² Diagnosis is based on patient history and physical examination and typical skin presentation. Image studies including CT, MRI, or lymphoscintigraphy can be arranged to assess severity and evaluate predisposing etiologies. Skin biopsy can be performed to rule out other differential diagnoses, including malignancy.

The goal of management is to investigate the underlying cause, correct associated risk factors, so as to prevent further skin complications. Conservative treatment includes the affected limb elevation, massage, and compression with bandages or stockings. Medical treatments include oral and topical retinoids, which decrease epidermal proliferation, fibrogenesis and inflammation. Surgical management are available, such as debridement, lymphovenous anastomosis and lymphatic transplantation.⁴

Conclusion:

In our case, long-term immobilization had predisposed higher risk of deep vein thrombosis formation. Without initial anticoagulation treatment, chronic venous thrombosis then induced venous insufficiency and lymphedema. Skin disfiguration with subsequent skin eruption also increased risk of infection, which led to the vicious cycle toward the formation of ENV.

It is important for physicians to understand and recognize the presentation of this uncommon condition. Especially in aging population with multiple comorbidities, long-term immobilization related chronic venous insufficiency would be a major risk factor of chronic lymphedema. Through recognizing the early presentation of lymphedema and correcting the possible underlying predisposing factors, physicians can prevent further dermatological complications.



Figure 1. Initial skin presentation

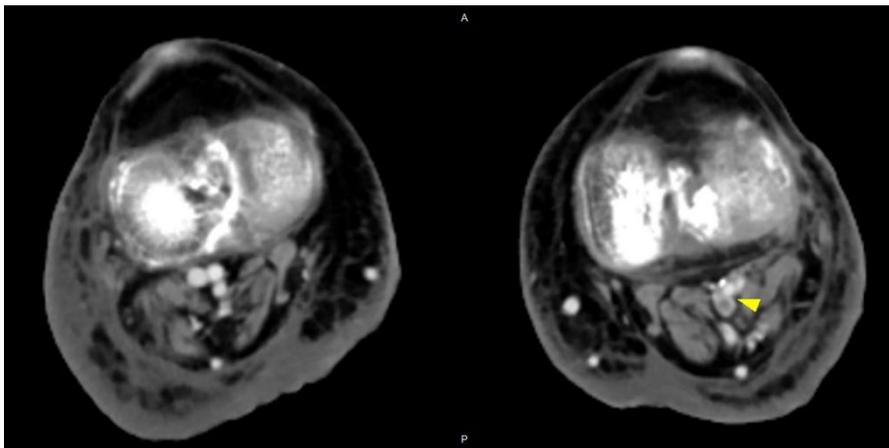


Figure 2. Thrombus in the left popliteal vein.



Figure 3. After one-week treatment

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