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Elephantiasis Nostras Verrucosa Secondary to Tuberculosis Verrucosa Cutis: A Rare Presentation of Cutaneous Tuberculosis

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Abstract:

Elephantiasis nostras verrucosa (ENV) is a rare form of chronic lymphedema, most commonly affecting the lower limbs. It has primary and secondary etiologies; primary lymphedema results from an intrinsic abnormality of the lymph-conducting pathway, while secondary lymphedema—which is more common—results from an acquired obstruction or obliteration of the lymph-conducting pathway. We report a 49-year-old female presenting with a slowly progressive verrucous swelling over the dorsum of the right lower leg for one year, clinically mimicking ENV. Histopathology revealed epithelioid granulomas with Langhans giant cells and caseous necrosis. Cartridge-based nucleic acid amplification testing (CBNAAT) confirmed Mycobacterium tuberculosis without rifampicin resistance, while the interferon-gamma release assay (IGRA) was positive. The patient was started on standard anti-tubercular therapy with early clinical improvement. This report describes a rare case of elephantiasis of the lower limb where the underlying etiology was cutaneous tuberculosis.

Keywords:

Tuberculosis verrucosa cutis, Elephantiasis nostras verrucosa, Chronic lymphedema, Cutaneous tuberculosis.

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Introduction

Elephantiasis nostras verrucosa (ENV) is a rare, exaggerated form of chronic nonfilarial lymphedema, most commonly affecting the lower limbs. Various factors can cause obstruction of the lymphatic system and result in ENV [1]. Chronic lymphedema has primary and secondary etiologies; primary lymphedema results from an intrinsic abnormality of the lymph-conducting pathway, and secondary lymphedema, which is more common, results from an acquired obstruction or obliteration of the lymph-conducting pathway. Congenital hypoplasia or agenesis is seen in Milroy's disease, resulting in primary lymphedema, while infections, malignancies, chronic venous stasis, post-radiation, trauma, obesity, and congestive heart failure lead to secondary lymphedema [2].

Tuberculosis verrucosa cutis (TVC) presents classically as thick, warty plaques over the extremities and sometimes over the buttocks. Involvement of the lymphatic system causing lymphadenopathy and lymphedema are known complications of tuberculosis infection. However, such a presentation in the setting of cutaneous tuberculosis is exceptionally rare [3].

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This case describes one such instance of elephantiasis of the lower limb where the underlying etiology was cutaneous tuberculosis.

Case Report

A 49-year-old female presented with a gradually progressive verrucous swelling over the dorsum of the right foot, which gradually involved the right lower leg over one year. The lesion initially started as a small papule following minor trauma while walking barefoot and progressively enlarged over time. There was no associated pain, itching, discharge, bleeding, or ulceration. The papule then spread to cover the entire dorsum of the right foot, gradually involving the lower leg up to the knee. No history of loss of appetite, fever, night sweats, cough, weight loss, or any other systemic symptoms was noted. Past, family, and personal histories were non-contributory. On examination, a few non-tender lymph nodes over the inguinal region were noted.

On dermatological examination, diffuse edema over the right foot and leg extending up to the knee was observed. A nodular hyperkeratotic verrucous plaque with irregular margins and focal crusting was noted over the dorsum of the right foot **(Figure 1)**. The lesion showed multiple hard nodular outgrowths with pseudo-papillary projections giving a cobblestone appearance over the shin. Stemmer's sign was positive.

Systemic examination was unremarkable. Routine hematological and biochemical investigations were within normal limits.
Histopathological examination **(Figure 2)** revealed acanthosis and hyperkeratosis of the overlying epidermis, along with the presence of an epithelioid granuloma in the dermis. High-power magnification demonstrated granulomas with surrounding fibroblastic proliferation, Langhans giant cells, and caseous necrosis.

CBNAAT (GeneXpert) from a tissue sample was positive for *Mycobacterium tuberculosis* with no rifampicin resistance detected. The interferon-gamma release assay (IGRA) was also positive. Based on the clinicopathological and molecular findings, a diagnosis of tuberculosis verrucosa cutis (TVC) presenting as ENV was established.
The patient was started on first-line anti-tubercular therapy (ATT) according to national guidelines. Follow-up evaluation demonstrated early clinical improvement with a reduction in hyperkeratosis and lesion thickness.

Discussion

In endemic regions, lymphatic filariasis remains the most common cause of lymphedema, and many patients are often empirically treated for filariasis irrespective of the underlying diagnosis. However, lymphedema can arise from many other causes. This case describes the occurrence of chronic lymphedema as a manifestation of tuberculosis, secondary to lymphangitis or lymphadenopathy leading to impaired lymphatic drainage and accumulation of lymph in the affected area. Despite this association, cutaneous tuberculosis presenting primarily with lymphedema has rarely been described in the literature [3, 4]. A similar case report was published by Sinha et al. where a 70-year-old male with ENV was later diagnosed to have secondary cutaneous tuberculosis [2].

The exact pathogenesis of ENV is not completely understood. It is believed that recurrent episodes of lymphangitis lead to progressive lymphatic damage, resulting in the leakage of protein-rich lymphatic fluid into the dermis and subcutaneous tissue. This initiates a chronic inflammatory cascade that initially causes pitting edema, which gradually progresses to nonpitting edema due to extensive fibrosis [1]. ENV then becomes non-pitting due to profound fibrosis, acquiring a woody hard consistency and a cobblestone appearance on the surface [2]. The Kaposi-Stemmer sign—the inability to pinch the skin on the dorsal aspect of the second toe—is a highly specific sign of chronic lymphedema, which was also positive in our case [7].

Impaired local immunity further predisposes the affected area to recurrent microbial infections, perpetuating the vicious cycle of inflammation and lymphatic damage. Tuberculosis verrucosa cutis is an exogenous form of cutaneous tuberculosis that commonly presents as slowly

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progressive verrucous plaques or nodules over trauma-prone sites, particularly the lower limbs. Although it usually responds well to ATT, untreated cases can rarely lead to secondary lymphedema and subsequently ENV [2].

The histopathology of TVC features prominent epidermal changes such as hyperkeratosis, acanthosis, and papillomatosis. Simultaneously, tuberculous granulomas with caseous necrosis of moderate intensity are seen in the dermis, and bacilli can sometimes be found [5, 6], which was consistent with our patient's histopathology. Further, CBNAAT (GeneXpert) from the tissue sample was positive for *M. tuberculosis* with no rifampicin resistance detected. IGRA was also positive, which has higher specificity and sensitivity compared to the tuberculin skin test (TST) [6].

Management of ENV involves two main approaches: treating the underlying cause and managing the lymphedema itself. In this case, the underlying TVC was treated with ATT (following chest and TB consultations), and supportive measures for ENV included limb elevation, physiotherapy, and pneumatic compression stockings to reduce symptoms. Diuretics and systemic retinoids may help decrease chronic edema. In severe, refractory cases, surgical debridement or even amputation may be required for recalcitrant cases [2, 8].

Conclusion

Although rare and scarcely reported in the literature, tuberculosis verrucosa cutis should be considered a possible underlying cause of elephantiasis nostras verrucosa in tuberculosis-endemic regions. This case highlights the importance of recognition through biopsy and molecular investigations, alongside early targeted treatment primarily aimed at halting disease progression, reducing lymphedema, and preventing further disability.

Declarations

Declaration of Patient Consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given consent for her clinical information and images to be reported in the journal. The patient understands that her name and initials will not be published and due efforts will be made to conceal her identity, although anonymity cannot be guaranteed.

Financial Support and Sponsorship: Nil.

Conflicts of Interest: There are no conflicts of interest.

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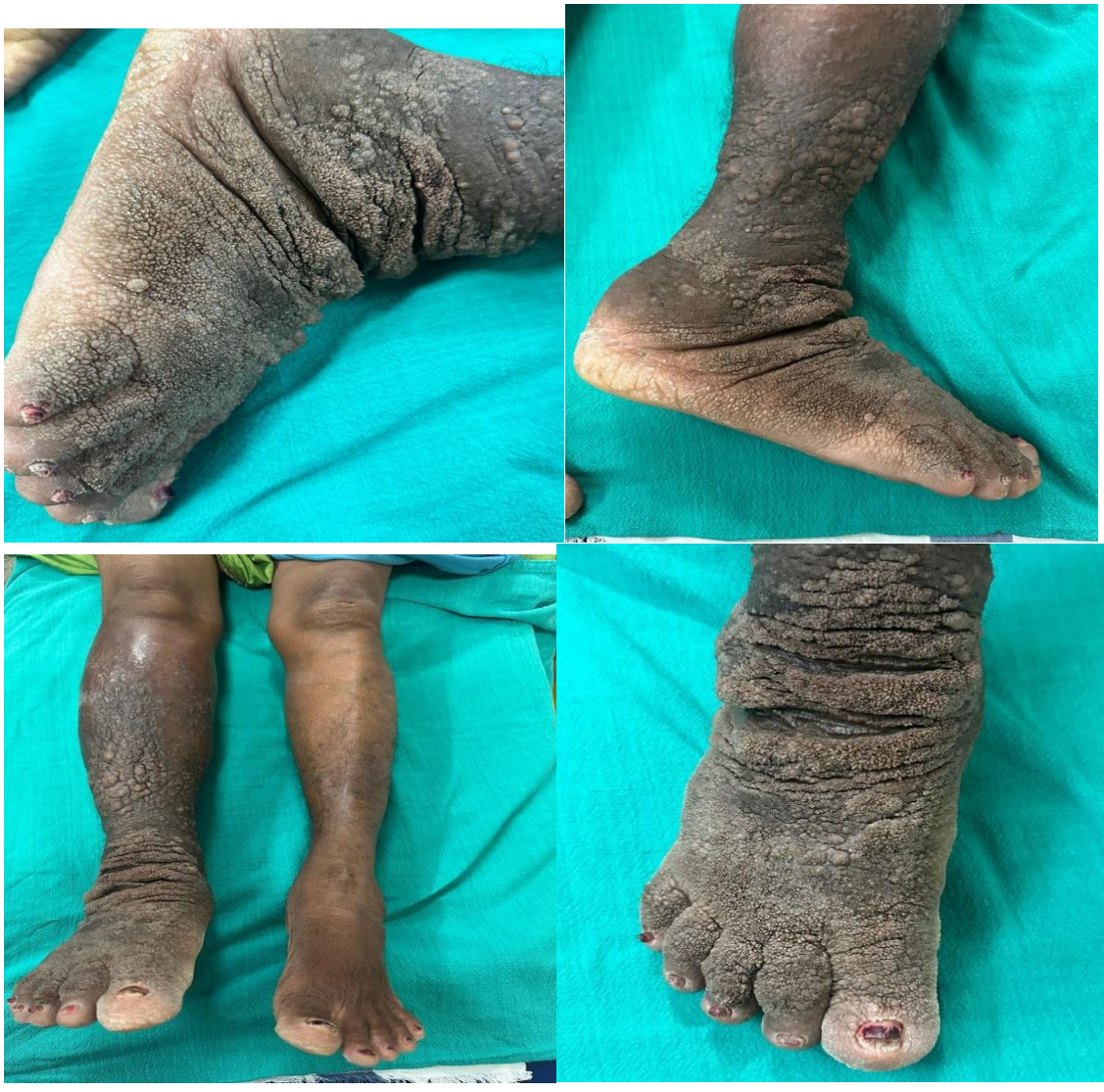
Figure Legends

(Note: Paste your images sequentially below these labels in your final document).

Figure 1 (A-E): Clinical presentation showing diffuse edema over the right foot and leg extending up to the knee. A nodular hyperkeratotic verrucous plaque with irregular margins, focal crusting, and pseudo-papillary growth giving a cobblestone appearance over the shin.



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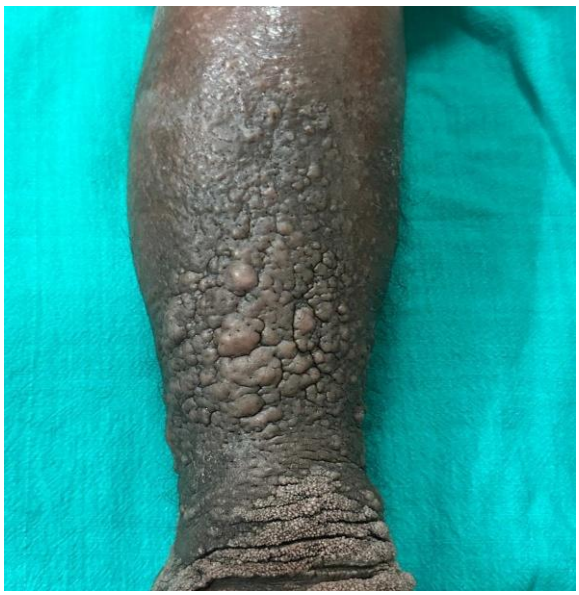
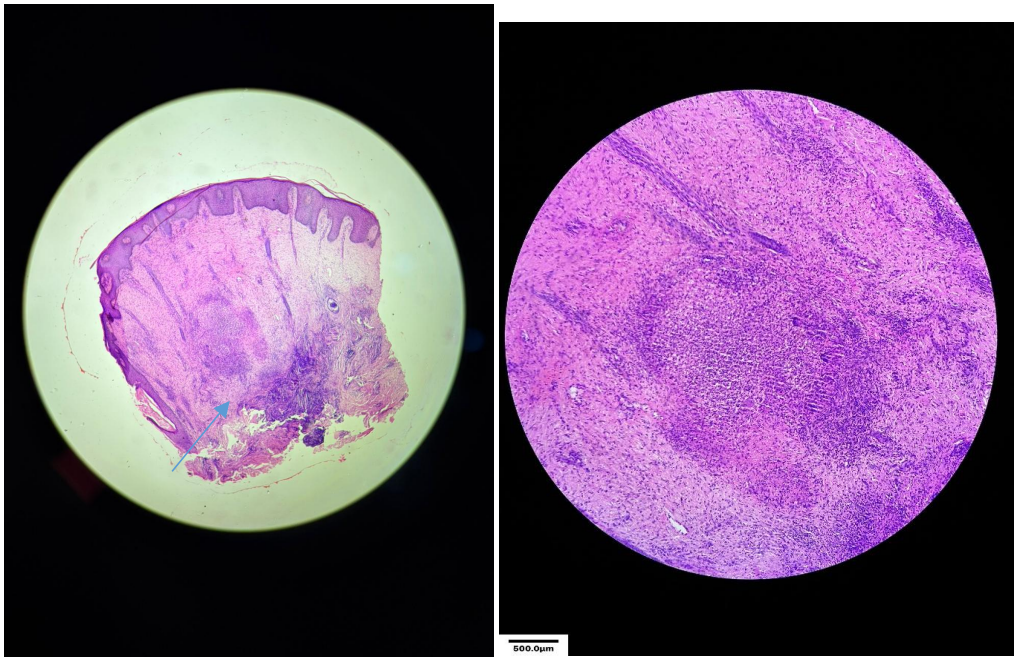


Figure 2 (A-C): Histopathological examination. (A) Low power view showing acanthosis and hyperkeratosis of the overlying epidermis, alongside an epithelioid granuloma in the dermis. (B) 10x view showing a granuloma with surrounding fibroblastic proliferation. (C) High power view highlighting Langhans giant cells and caseous necrosis.



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