Case Presentation

Reactive eccrine syringofibroadenoma in an elderly patient

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Abstract

Eccrine syringofibroadenoma (ESFA) is an uncommon benign adnexal tumor with eccrine differentiation, with fewer than 100 cases reported worldwide. Since its initial description, several clinical subtypes have been recognized, including a reactive form associated with chronic inflammatory or neoplastic conditions. We report the case of an 88-year-old man with extensive reactive ESFA developing in the setting of long-standing postthrombotic lymphedema and neglected ulceration. The patient presented with poor general health, bilateral pleural effusions, multiple verrucous nodules, severe stasis ulcers, and secondary polymicrobial and fungal infections. Histopathologic examination of a nodule demonstrated interconnecting cords of cuboidal epithelial cells with ductal structures within a fibrovascular stroma, consistent with eccrine differentiation and diagnostic of ESFA. Management focused on infection control, wound care, and supportive therapy, as surgical excision and amputation were not suitable options. Clinical improvement was achieved; however, ongoing monitoring was advised because of the potential for malignant transformation. This case highlights the role of chronic tissue injury in the pathogenesis of reactive ESFA and underscores the importance of histopathology for diagnosis, multidisciplinary collaboration in management, and individualized treatment planning in elderly patients with advanced comorbidities.

Introduction

Eccrine syringofibroadenoma (ESFA) is a benign cutaneous tumor with eccrine differentiation, with approximately 80 cases reported worldwide.[1] Mascaró[2] first described ESFA in 1963 as a solitary, benign tumor with distinctive histopathologic features. In 1997, Starink[3] classified ESFA into 4 subtypes based on clinical presentation and associated conditions: (1) solitary ESFA, presenting as a single lesion, most often on the extremities, without systemic association; (2) multiple ESFA associated with hidrotic ectodermal dysplasia; (3) multiple ESFA without associated cutaneous findings; and (4) nevoid ESFA, characterized by nonfamilial, unilateral lesions that are often congenital or of early onset. Subsequently, a fifth subtype (reactive ESFA) was proposed, associated with chronic inflammatory or neoplastic conditions.[4]

Case Synopsis

We present the case of an 88-year-old man who presented to our clinic in poor general health. His medical history was significant for atrial fibrillation and postthrombotic syndrome. His condition had progressed over many years, with clear evidence of neglect, as this was the first time he sought medical attention. Physical examination revealed bilateral pleural effusions and numerous verrucous nodules on both legs, predominantly on the left (Figure 1). The left leg also demonstrated marked edema and stasis ulcers. The ulcers were malodorous and showed signs of secondary infection. Microbiologic analysis of ulcer cultures revealed a polymicrobial infection, with *Staphylococcus aureus* and *Escherichia coli* identified. In addition, fungal cultures from skin scales confirmed the presence of *Candida albicans*. Laboratory evaluation demonstrated hypoalbuminemia and electrolyte imbalance, consistent with chronic disease.

A biopsy of a nodule on the left foot was performed, and histopathologic examination was undertaken to determine the etiology. Histologic analysis demonstrated a lattice-like arrangement of thin cords of cuboidal epithelial cells within a fibrovascular stroma. These epithelial cords exhibited ductal structures indicative of eccrine differentiation and were connected to the epidermis. Additional findings included verrucous epidermal hyperpla-

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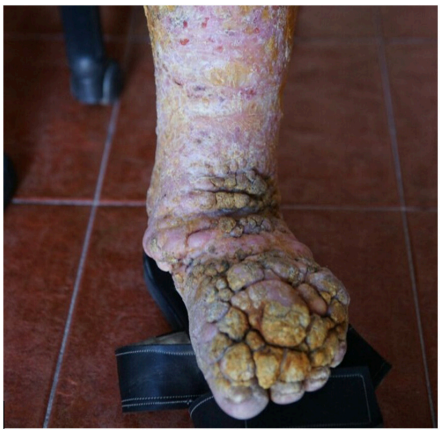

Figure 1. *Clinical presentation showing multiple verrucous nodules on the left foot.*

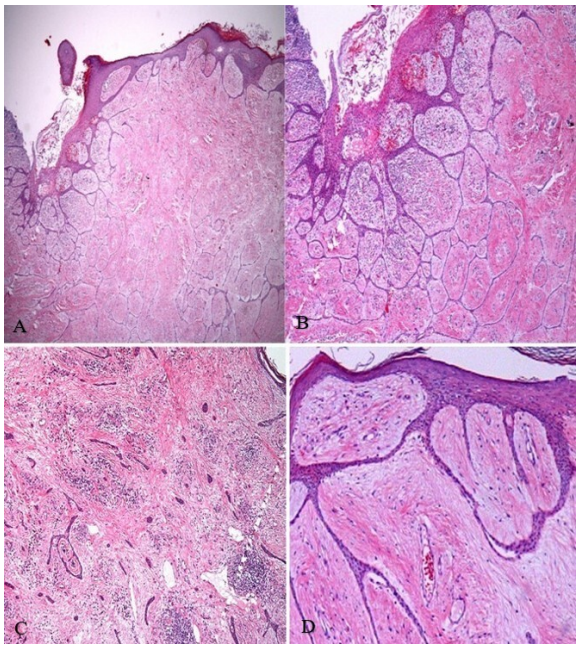

Figure 2. *Histopathologic findings.* ***(A)*** *Verrucous epidermal proliferation with thin, anastomosing cords and strands of epithelial cells extending from the epidermis into the dermis within a fibrovascular stroma (hematoxylin-eosin, original magnification ×4).* ***(B)*** *Eccrine hyperplasia with a reticulated pattern (hematoxylin-eosin, original magnification ×10).* ***(C)*** *Hyperplasia of eccrine ducts (hematoxylin-eosin, original magnification ×10).* ***(D)*** *Ductal differentiation within epithelial strands (hematoxylin-eosin, original magnification ×40).*

sia, dermal fibrosis, and hamartomatous vascular structures (Figure 2). These features were consistent with a diagnosis of ESFA.

Case Discussion

This case describes a patient with ESFA arising in the setting of chronic postthrombotic lymphedema and long-standing ulceration. Reactive ESFA typically affects individuals in the sixth to eighth decades of life, with no clear sex predilection. Its pathogenesis is thought to involve chronic tissue injury and repair, resulting in hyperplastic and hamartomatous proliferation of eccrine ducts. Conditions such as diabetes mellitus, autonomic dysfunction, and neuropathy can lead to recurrent trauma, ulceration, and impaired wound healing, thereby promoting ductal hyperplasia. Similar mechanisms have been described in leprosy, burn scars, and chronic inflammatory dermatoses. Altered sympathetic nerve function has also been implicated in aberrant eccrine regeneration, further contributing to the reactive process.[1,4,5]

The diagnosis of ESFA relies on its characteristic histopathologic features, which are consistent across all subtypes. Microscopically, ESFA is characterized by interlacing cords and strands of epithelial cells forming duct-like structures within a fibrovascular stroma that often contains an inflammatory infiltrate with plasma cells. Because this pattern may represent a reactive process, it is essential to exclude underlying inflammatory dermatoses, adjacent malignancy, or malignant transformation.[6]

Management was multidisciplinary and included systemic antibiotics and antifungal therapy, cardiovascular management, and supportive care, including fresh frozen plasma, whole blood transfusions, and electrolyte replacement to correct underlying deficiencies. Local wound care consisted of regular cleansing and debridement to manage stasis ulcers and prevent secondary infection. This comprehensive approach resulted in clinical improvement and stabilization of the patient's condition. Management strategies for ESFA depend on subtype, clinical presentation, and patient preference and may include observation, surgical excision, CO_2 laser ablation, or radiotherapy.[5,7] In this case, surgical excision was not feasible because of the extensive number of nodules, and the patient declined lower limb amputation. Regular follow-up was recommended to monitor for potential malignant transformation.

Conclusion

Reactive ESFA may develop in the setting of long-standing lymphedema, ulceration, and recurrent infection, in which chronic tissue injury gives rise to rare adnexal proliferations. Histopathologic examination is essential for accurate diagnosis, as clinical features may mimic those of other benign or malignant tumors. Although malignant transformation is uncommon, ongoing surveillance is warranted. In elderly patients with multiple comorbidities, a multidisciplinary approach is necessary to achieve optimal outcomes.

Potential conflicts of interest

The authors declare no conflicts of interest.

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