CASE REPORT

The story of a deformed leprous foot

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Summary Eccrine syringofibroadenoma (ESFA) is a rare adnexal tumour of eccrine ductal proliferation. A 50 year old treated case of leprosy presented with a chronic non healing ulcer of 5 years duration on the deformity laden right foot. Multiple verrucous papules and plaques were seen surrounding the ulcer which showed histopathological findings consistent with ESFA. Although ESFA constitutes a rare association with leprosy, considering the load of treated cases in our country and elsewhere, it may represent an under-reported entity which requires more attention in the post elimination era.
Introduction

Eccrine syringofibroadenoma (ESFA) is a rare adnexal tumour of eccrine ductal proliferation with variable clinical findings and characteristic histological features. This entity has been well described in literature, but the reactive subtype of ESFA has generated renewed interest. Increasingly, newer associations of this emerging subtype with inflammatory dermatoses are being reported and tissue remodelling has been suggested as the purported mechanism for its occurrence.\(^1\) We report a patient with chronic non healing ulcer on a deformed leprous foot with surrounding areas showing changes consistent with ESFA.

Case report

A 50 year old female presented with an ulcer over the sole of her right foot since 5 years. The patient gave a history of decreased sensation over the right foot and was a known case of lepromatous leprosy released from treatment 3 years back after completing multibacillary multi-drug therapy for 2 years. She had been treated intermittently with topical and systemic antibiotics in addition to daily dressings of the ulcer, but complete resolution was never realised.

Cutaneous examination revealed a fixed flexion and adduction deformity of the right foot, along with flexion deformity of the right great and second toe. Examination revealed a single localised non healing ulcer measuring 7 × 5 × 2 cm over the plantar aspect of the right foot with sloping edges, indurated margin and pale granulation tissue with seropurulent discharge at the base (Figure 1).

Figure 1. Multiple verrucous papules and plaques (arrows) surrounding the ulcer on the right foot.
In addition, multiple verrucous papules and plaques were seen surrounding the ulcer and also extending onto the lateral and dorsal surface of the foot (Figure 1).

The right leg showed ichthyotic changes along with hyperpigmentation. Touch and pain sensation were lost over the foot and up to the lower one-third of the right leg. Nerve thickening and regional lymphadenopathy were absent and systemic examination was non-contributory. The differentials thought of were squamous cell carcinoma (SCC), tuberculosis verrucosa cutis and chromoblastomycosis.

The routine laboratory examination and serum biochemistry panel were normal. A Mantoux test was negative. KOH mount from the ulcer showed no fungal elements. X-ray of the right foot showed destruction and dislocation of talus, metatarsal and phalanges along with increased density of the bones and mild soft tissue swelling. The features were suggestive of neuropathic joints. Culture from the ulcer showed growth of *Pseudomonas aeruginosa*, whereas fungal culture was negative. A wedge biopsy from the verrucous growth showed slender anatomising epithelial cords of acrosyringeal cells with formation of lumina embedded in fibrovascular stroma (Figures 2a, b, c).

Based on clinical findings and histopathology, a diagnosis of trophic ulcer with ESFA in a treated case of leprosy was made. The patient was treated with systemic antibiotics and daily saline dressings. The ulcer has partially resolved and as the patient refused amputation, we are keeping her under observation. Due to a large area of involvement and trophic changes in the foot, excision of the tumour was not attempted.

**Discussion**

Reported for the first time by Mascaro, ESFA is a rare benign cutaneous adnexal lesion that predominantly occurs in patients over 40 years of age. It has a strikingly polymorphous...
clinical presentation ranging from a solitary papule or nodule to multiple lesions with a linear or papulonodular distribution.\textsuperscript{3} Despite the variability of clinical presentation, ESFA is histologically unique. It is a distinct tumour composed of a proliferation of anastomosing cords of monomorphous epithelial cells harbouring eccrine ductal formations all admixed within an inflammatory fibrovascular stroma.\textsuperscript{3}

Starink\textsuperscript{4} classified the condition into four types: solitary, multiple and associated with ectodermal dysplasia, multiple without associated cutaneous findings and non-familial unilateral linear (sometimes referred to as nevoid ESFA). In addition to these, French\textsuperscript{5} added

\begin{figure}[h]
\centering
\includegraphics[width=0.8\textwidth]{figure2b.png}
\caption{Higher power of the same sections showing several ductal structures within the anastomosing epithelium (black arrow). The stroma (blue arrow) is better seen at this power of magnification (H & E, $\times$100).}
\end{figure}

\begin{figure}[h]
\centering
\includegraphics[width=0.8\textwidth]{figure2c.png}
\caption{High power view showing a duct (black arrow) and mucin in the adjacent stroma (blue arrow) (H & E, $\times$400).}
\end{figure}
a fifth reactive subtype to inculcate all reports of ESFA associated with inflammatory or neoplastic dermatoses and also those resulting due to peripheral neuropathy.

The pathogenesis of reactive ESFA is uncertain, and it has been suggested that it may result from repeated eccrine duct trauma resulting in eccrine duct remodeling and repair.\textsuperscript{5,6} Apart from leprosy, reactive ESFA has been reported in other conditions causing peripheral neuropathy such as diabetes mellitus.\textsuperscript{7} The insensitive feet due to peripheral neuropathy make it prone for recurrent traumatic events resulting in non healing wounds and ulcers. Proliferation of sweat duct epithelium in scar areas and in re-epithelialisation of healing wounds is reported to be a common response.\textsuperscript{8} The possibility of a neuroeccrine interaction, with sympathetic neuropathy in leprosy as a contributing factor in the pathogenesis has also been considered.\textsuperscript{9}

Although ESFA usually follows a benign course, few authors have reported malignant transformation into SCC and porocarcinoma.\textsuperscript{10,11} But malignancy occurring in reactive ESFA developing in the context of inflammatory dermatoses has not been reported. Therefore, a wait-and-see approach is a reasonable option especially in leprosy associated ESFA where trophic changes compromise the wound healing capacity of the patient.\textsuperscript{12}

To the best of our knowledge, leprosy associated reactive ESFA has only been reported on two prior occasions.\textsuperscript{6,9} Although the prevalence of leprosy is on the decline, burnt out cases like this patient who do not have active disease but continue to face the neurological sequelae of leprosy are numerous. We highlight this case to propagate our view that leprosy associated ESFA may be an under-reported entity. Considering the large number of patients who have completed treatment and suffer from trophic changes in the lower extremities, we advocate careful screening of all such patients to rule out ESFA.

References