Syringofibroadenoma Arising Adjacent to Verrucous Carcinoma: Is It a Reactive Process, Malignant Transformation, or Collision Tumor?

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Abstract

Eccrine syringofibroadenoma (ESFA) is a rare, benign tumor showing acrosyringeal or sweat gland differentiation with variable clinical presentations, but distinct histopathological features. Rarely, ESFA coexists with a malignancy such as a squamous cell carcinoma or porocarcinoma. Herein, we report the first case of an ESFA associated with a verrucous carcinoma in a 46-year-old female on the right foot. It is unclear whether it represents a reactive process, a malignant transformation or a collision tumor. We recommend ESFA be completely excised if possible or closely followed given its association with a developing carcinoma.

Keywords: Eccrine syringofibroadenoma, reactive, verrucous carcinoma

INTRODUCTION

Eccrine syringofibroadenoma (ESFA) is a rare, benign tumor showing acrosyringeal or sweat gland differentiation, with variable clinical presentations and distinct histopathological features.¹ It usually occurs in the extremities of old individuals as a slow-growing verrucous nodule or plaque.²⁻⁵

Five clinically distinct subgroups have been proposed and include: (1) solitary ESFA, (2) multiple ESFA associated with ectodermal dysplasia, (3) multiple ESFA without associated cutaneous findings, (4) nonfamilial unilateral linear ESFA, and (5) reactive ESFA.⁶ Reactive type ESFA has been described to coexist with various inflammatory processes including ulcers and venous stasis, as well as neoplastic conditions such as squamous cell carcinoma (SCC), basal cell carcinoma (BCC), eccrine poroma, and hidroacanthoma simplex. Herein, we report the first case of an ESFA associated with verrucous carcinoma.⁷⁻⁸

CASE REPORT

A 46-year-old female with a medical history of poorly controlled, insulin-dependent Type 1 diabetes mellitus and hypertension, presented with a friable pigmented verrucoid lesion present for many years on the posterior aspect of the right calcaneus [Figure 1a]. A second flatter and nonfriable lesion connected to the verrucoid lesion and extended on to the lateral and dorsal aspect of the foot, measuring 7 cm × 3 cm in size [Figure 1b]. Both of the lesions were nonpainful and did not show signs of infection. The differential diagnosis included plantar keratoderma and verrucous carcinoma.

An excision of the verrucoid lesion was obtained from the right calcaneus. Histologic examination revealed an acanthotic papillary proliferation showing massive hyperkeratosis and parakeratosis. The well-differentiated squamous epithelium invaded into the dermis with bulbous processes and a “pushing” border consistent with verrucous carcinoma [Figure 2]. Due to the patient’s poor glycemic control, she was not considered to be an appropriate candidate for surgery. Six months later, after the diabetes was better controlled, she underwent an excision of the second lesion on the lateral and dorsal aspect of the foot.

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Microscopic examination revealed a broad epidermal proliferation with anastomosing strands of monomorphic round cells which hang from the overlying epidermis, enclosing a fibrovascular stroma with a prominent lymphoplasmacytic infiltrate. The epithelial strands showed ductal differentiation consistent with an ESFA [Figure 3]. There were no features of malignancy identified.

**DISCUSSION**

ESFA also called an acrosyringeal nevus, is a rare benign adnexal tumor which was first described by Mascaro in 1963. Clinically, it manifests as solitary or multiple hyperkeratotic plaques or papulonodules in a diffuse, linear or clustered distribution. The anatomical location and age of onset can be quite variable, depending on the clinical subtype, but are most common on the extremities of elderly patients.

Reactive ESFA has been seen in association with different inflammatory diseases including diabetic foot ulcer, vascular stasis, lichen planus, bullous pemphigoid, epidermolysis bullosa, primary cutaneous amyloidosis, burn scar, hyperkeratotic eczema, pincer nail, ileostomy stoma, plantar hyperkeratosis, leprosy and various neoplastic conditions such as SCC, BCC, eccrine poroma or porocarcinoma and hidroacanthoma simplex. The pathogenesis of reactive ESFA is poorly understood but is considered by some to be a reactive epithelial proliferation instead of a neoplasm of eccrine glands as spontaneous regression has been described after successful treatment of the associated underlying inflammatory condition.

Although the clinical course of ESFA is typically benign, malignant transformation to eccrine syringofibrocarcinoma and its association with malignancies such as SCC, porocarcinoma, and currently verrucous carcinoma is concerning. It is difficult to determine whether ESFA is a reactive process, a precursor to carcinoma, or a benign tumor that arises concurrently.

In the current patient as the ESFA and the verrucous carcinoma were excised separately and 6 months apart we do not have any sections containing both entities together. However, on the clinical examination, the two lesions appeared adjacent to each other. Neither was surrounded by nor intermingled with the other one. Therefore, it is not clear if the verrucous carcinoma is a malignant transformation of the ESFA.

Since association of ESFA and diabetes mellitus and the proposed hypothesis for the responsible physiopathology have been previously reported and discussed, in the current case, patient’s known diabetes mellitus could be an underlying contributory factor although no diabetic ulcer was present at the location of the ESFA and the presence of neuropathy was not documented in the patient.

Whether the underlying diabetes and verrucous carcinoma are the perpetrators for inducing ESFA or if their occurrence is just coincidental is not known.

The mainstay of treatment is surgical excision for solitary ESFA given malignant transformation has been reported. However, the risk appears to be low, and observation with close clinical follow-up may be an adequate alternative, after judicious sampling, especially with larger lesions not amenable to surgery.

To the best of our knowledge, the association of reactive ESFA with verrucous carcinoma has not been previously reported. Whether it is a so-called collision tumor, a reactive response or precursor to verrucous carcinoma is debatable. The biological behavior and pathophysiology of these lesions warrant further evaluation of similar cases.

![Figure 1: Gross photographs of the (a) verrucoid lesion on the right calcaneus and (b) second nonfriable lesion on the dorsolateral aspect of the foot](image1)

![Figure 2: Verrucous carcinoma. Broad papillomatous acanthotic blunted projections with overlying marked hyperkeratosis, parakeratosis, and hypergranulosis. The underlying fibrous stroma contains ectatic vessels and a mixed chronic inflammatory infiltrate (×40)](image2)

![Figure 3: Syringofibroadenoma. (a) Broad epidermal proliferation with anastomosing strands (×20), comprised of (b) small uniform cells with ductal differentiation and no cytological atypia (×100)](image3)
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Conflicts of interest
There are no conflicts of interest.

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