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REACTIVE SYRINGOFIBROADENOMATOUS HYPERPLASIA IN CHRONIC PLAQUE PSORIASIS: A RARE CASE REPORT.

Dermatology

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ABSTRACT

Eccrine syringofibroadenomatous hyperplasia is a rare benign cutaneous adnexal lesion characterized by a hyperplastic epithelium and eccrine ductal differentiation. In the present case, a 57-year-old male presented with numerous, well defined erythematous to hyperpigmented hyperkeratotic plaques with warty nodules on surface. On histopathological examination, diagnosis of eccrine syringofibroadenomatous hyperplasia was confirmed. Our unusual and interesting case report emphasizes a case in which multiple cobblestone like eccrine syringofibroadenomatous hyperplastic lesions arise from chronic plaque psoriasis.

KEYWORDS

Syringofibroadenomatous Hyperplasia, Psoriasis, Eccrine

INTRODUCTION:

We report a case of chronic plaque psoriasis developing acuminate growth clinically, which revealed syringofibroadenomatous hyperplasia on histological examination.

CASE REPORT:

57 year old male patient presented with asymptomatic lesions over both legs, hands and trunk since 10 years which gradually increased in size and became warty since 2 years. There was history of application of topical steroid. On cutaneous examination there were well defined erythematous to hyperpigmented hyperkeratotic plaques with and warty nodules on surface.

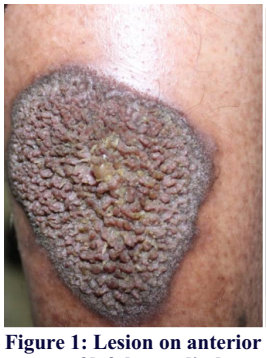

Figure 1: Lesion on anterior aspect of left lower limb

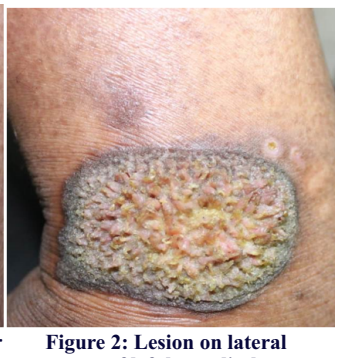

Figure 2: Lesion on lateral aspect of left lower limb

Clinical diagnosis was chronic plaque psoriasis with acuminate growth. General examination and systemic examination was normal. Complete hemogram, liver and renal function tests were normal. Skin biopsy was done and histopathological examination revealed papillomatosis of epidermis with orthohyperkeratotic stratum corneum with hypergranulosis and focal areas of parakeratosis. Multiple eccrine ducts were noted.

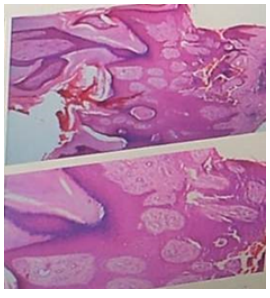

Figure 3 Histopathological section of lesion on left lower limb

The impression was reactive syringofibroadenomatus hyperplasia in chronic plaque psoriasis.

DISCUSSION:

Eccrine syringofibroadenomatous hyperplasia, also called eccrine syringofibroadenoma (ESFA) is a rare benign adnexal tumor arising most often on the extremities of elderly individuals with characteristic histopathology. The tumor consists of anastomosing cords of cuboidal epithelial cells surrounded by a fibrovascular stroma containing plasma cells and ductal structures[1]. Although the exact site of origin of ESFA remains controversial, it is believed to be derived from the acrosyringium or eccrine dermal duct. ESFA stains positively with epithelial membrane antigen (EMA) and carcinoembryonic antigen (CEA). Cases of ESFA have been divided into five distinct subtypes due to clinical manifestations: solitary lesions, multiple lesions associated with an ectodermal dysplasia{Schopf syndrome}, lesions with no additional cutaneous pathology, nevoid lesions, and reactive lesions.

1. Solitary ESFA is typically a nonhereditary solitary nodule or verrucous mass normally found on the lower extremities of the middle-aged and elderly.
2. Multiple ESFA without associated cutaneous finding (eccrine syringofibroadenomatosis) are nonfamilial palmoplantar lesions without significant associated cutaneous finding.
3. Multiple ESFA with hidrotic ectodermal dysplasia (HED) presents in two different variants, Schöpf-Schulz-Passarge syndrome and Clouston's syndrome. Schöpf-Schulz-Passarge syndrome (SSPS) is a rare autosomal dominant ectodermal dysplasia, characterized by hypodontia, hypotrichosis, nail dystrophy, palmoplantar keratoderma, and periocular and eyelid margin apocrine hidrocystoma. Clouston's syndrome is autosomal dominant ectodermal dysplasia, characterized by dystrophic nails and sparse hair. HPV-10 has been detected in the lesions occurring in Clouston's syndrome[2].
4. Nevoid ESFA (nonfamilial unilateral linear ESFA) is a rare genetic mosaicism, producing diffuse plantar hyperkeratosis. It may be the result of a somatic mutation in the early embryonic stage
5. Reactive ESFA associated with inflammatory or neoplastic dermatosis is often in an acral location. This reactive change has been seen next to burn scar ulcer, erosive palmoplantar lichen planus[3], hidrotic ectodermal dysplasia, bullous pemphigoid[4], diabetes mellitus with polyneuropathy and chronic ulcer, inflammatory psoriasis or persistent local infection, lymphedema (elephantiasis), nevus sebaceous[5] and squamous cell carcinoma. The pathogenesis may be associated with a specific type of eccrine duct remodeling or repair. Inflammation due to repetitive trauma or local inflammatory disease would trigger syringomatous proliferation.

In immunohistochemical studies cytokeratin expression has stressed pathogenic role of dysregulated differentiation[6]. Epithelium undergoes differentiation towards eccrine excretory portion. Saggini and Mully[7] suggested that Pleckstrin homology-like domain, family A, member 1 protein (PHLDA-1), an anti-apoptotic protein expressed by hair follicle bulge keratinocytes, is expressed by the basal layer of ESFA strands and cords. Increased level of PHLDA-1 has been demonstrated in the hyperplastic epithelial networks of fibroepithelioma of Pinkus (which resembles that of ESFA) and the basal layer of the junction between the acrosyringium and eccrine dermal duct.[8]

Malignant transformation of ESFA has been noted[9,10]. In the reported cases, the following unifying features were identified: (1) patients age (averagely seventh or eighth decade), (2) male patients, (3) involving the extremities, and (4) concerned clinical manifestation including new growth, ulceration and crusting, and persistent disease despite extensive treatment. There are no reports of malignancy occurring in the setting of reactive ESFA developing from inflammatory dermatoses.

The treatment of ESFA relies on the number, area, location and resectability of the lesions. Solitary ESFA could be completely cured by surgical excision[11]. Additionally, complete excision seems to be the appropriate treatment for ESFA associated with malignant risk in the limited reports available. If skin lesions are difficult to be excised, multiple biopsy is necessary to exclude malignant transformation. Spontaneous regression of reactive ESFA after successful treatment of the underlying inflammatory condition has been described

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