Eccrine syringofibroadenoma: a case report and review of literature.

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ABSTRACT:

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Eccrine syringofibroadenoma (ESFA) is a rare, benign tumor of eccrine derivation which has variable clinical presentation and characteristic histology. It usually presents on extremities of elderly patient. ESFA has been classified into 5 different subtypes. Histologically,characterized by proliferation of cords and strands with ductal differentiation embedded in fibrovascular stroma. Clinical course is typically benign, however malignant transformation has been reported. In this report, we demonstrated a patient who presented with multiple hyperkeratotic verrucous papules for 10 years and histology compatible with ESFA.

Key word: Eccrine syringofibroadenoma, eccrine tumor

บทคัดย่อ:

ปุณยพัศฐิช์ ศิริธนบดีกุล พลอยทราย รัตนเขมากร รายงานผู้ป่วยเนื้องอกท่อต่อมเหงื่อ และทบทวนบทความทางวิชาการ. วารสารโรคผิวหนัง 2554; 27: 334-337.

หน่วยโรคผิวหนัง ภาควิชาอายุรศาสตร์ คณะแพทยศาสตร์ โรงพยาบาลรามาธิบดี มหาวิทยาลัยมหิดล

Eccrine syringofibroadenoma (ESFA) เป็นโรคเนื้องอกที่เจริญมาจากต่อมเหงื่อซึ่งพบได้ไม่บ่อย มีลักษณะทางคลินิกได้หลากหลายแต่ลักษณะทางพยาธิวิทยาค่อนข้างจำเพาะและช่วยในการวินิจฉัยโรค โรคนี้มักพบบริเวณขาโดยเฉพาะมือและเท้าในผู้สูงอายุ ส่วนใหญ่การดำเนินโรคไม่รุนแรง อย่างไรก็ตามมีรายงานการเปลี่ยนแปลงเป็นมะเร็งได้

รายงานฉบับนี้เป็นการนำเสนอผู้ป่วยชายอายุ 68 ปี มาตรวจเนื่องจากมีผื่นนูนที่เท้าทั้ง 2 ข้างนาน 10 ปี ลักษณะทางจุลพยาธิวิทยาเข้าได้กับ Eccrine syringofibroadenoma

Key word: เนื้องอกท่อต่อมเหงื่อ

Case report

A 68-year-old Thai man from Chaiyaphum presented with 10 years duration consisting of the eruption of dry hyperkerototic verrucous plaques involving both feet with occasionally itch. He was treated with oral and topical medications for period of times from primary physician without any improvement. His past medical history was chronic inactive hepatitis B infection which required no treatment. Dermatologic examination revealed multiple dry hyperkeratotic verrucous nodules and plaques markedly on lateral side of left foot and also less occur on lateral side of right foot (Figure 1,2). There was no other abnormal systemic symptom. There was no family history of similar skin condition. A punch biopsy obtained from his lesion and stain with hematoxylin-eosin. Histopathological finding showed thin elongated anastomosing cords of uniform small epithelial cells, emanating from the epidermis and extending into fibrongiomatous stroma with compact hyperkeratosis, hypergranulosis, and epidermal hyperplasia (Figure 3,4). With the clinical and histopathological finding, the diagnosis of eccrine syringofibroadenoma was made. Unfortunately, patient loss to follow up since then.

Figure 1

Figure 2

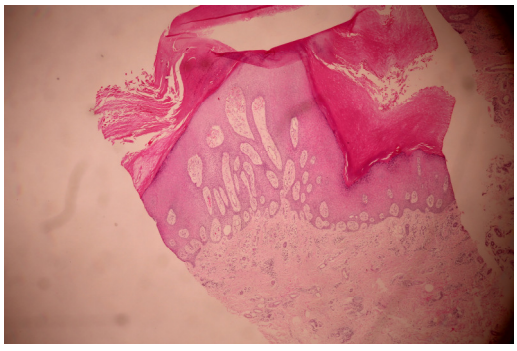

Figure 3

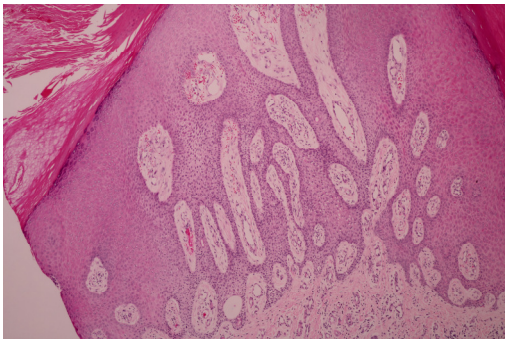

Figure 4

Discussion:

Eccrine syringofibroadenoma (ESFA) is an uncommon tumor of eccrine derivation that was first described by Mascaro in 1963[1] as a skin tumor with a histopathological picture mimic fibroadenoma of the breast and fibroepithelial type of basal cell carcinoma[2].

ESFA typically presents as a slow-growing, solitary or multiple, flesh- or reddish-colored nodule or plaque, with a predilection for the extremities especially palms and soles, however other sites such as faces, back of hands, nail and lumbar area have been reported[3]. It usually affects eldery patient with most presenting in their seventh and eighth decades[4]. Recently, EFSA has been classified according to the clinical presentation into 5 subtypes[5] including 1) Solitary ESFA, 2) Multiple ESFA in the SchÖpf syndrome 3)Multiple ESFA without associated cutaneous findings, 4) Non-familial unilateral linear ESFA, 5) Reactive ESFA (Table1)

Table 1 Classification of EFSA according to clinical presentation

Classification	**Description**
Solitary EFSA	Most common type. Non hereditary, verrucous growth, occur in middle-aged and elderly patients
Multiple ESFA in the SchÖpf syndrome	Hereditary; multiple erythematous papules in mosaic pattern on the palms and soles associated with hydrocystoma, hypotrichosis, hypodontia and nail dystrophy
Multiple ESFA without associated cutaneous findings	Non hereditary, palmoplantar lesion, no associated cutaneous finding
Non-familial unilateral linear ESFA	Rare, probably represent a genetic mosaic caused by a postsomatic mutation
Reactive ESFA	Associated with inflammatory dermatoses and blistering disorder such as bullous pemphigoid, lichen planus and burn scar, etc.

EFSA are believed to arise from acrosyringium, dermal sweat duct or both. The histological appearance characterized by proliferation of anastomosing cords and strands of cuboidal epithelial cells in a reticular pattern extending from epidermis into dermis. The strands contain tubular structure with eccrine duct formation embedded in a fibrovascular stroma with lymphocytes and plasma cells[6]. The histologic differential diagnosis includes fibroepithelia tumor of Pinkus, tumor of the follicular infundibulum pseudoepitheliomatous hyperplasia, papillary eccrine adenoma, reticulated seborrheic keratosis, squamous cell carcinoma, and artifacts of histologic processing[1].

ESFA is typically benign clinical course. Unfortunately, recent reports demonstrate the possibility of malignant transformation to eccrine syringofibrocarcinoma and the co-existence with squamous cell carcinoma has been reported in cases of ESFA. HPV type 10-DNA has been reported in a case of syringofibroadenoma which may play a role in malignant transformation. However, whether the ESFA develop in response to a SCC or the SCC represents a malignant degeneration of the ESFA is still unclear[6]. The worrisome features including increase in size, pain, inflammation, new growth, ulceration and crusting, and persistent disease despite extensive treatment have been associated with malignancy changes[4].

The treatment of ESFA depends on the number, location, and resectability of lesions. Excision is the mainstay of treatment for solitary ESFA, however if tumor is unresectable, generous sampling to rule out malignant transformation is suggested.

Dual pulse width flashlamp pulsed dye laser, CO2 laser and radiation therapy may be alternative treatment options which have been reported to be an effective treatment especially for large lesion or located at difficult anatomical site[3, 7, 8].

In case of ESFA associated with or exhibiting malignant feature, complete excision is the appropriate treatment. In contrast to reactive type, the risk of malignant transformation is quite low and spontaneous involution has been reported, thus monitoring instead of excision is a reasonable option.[9]

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