Solitary eccrine syringofibroadenoma: a case report showing papillary tubular adenoma-like features

Toshiyuki Yamamoto¹, Keiko Miura², Hiroo Yokozeki¹

¹Department of Dermatology, Tokyo Medical and Dental University, 1-5-45 Yushima, Tokyo 113-8519, Japan, ²Department of Pathology, Tokyo Medical and Dental University, 1-5-45 Yushima, Tokyo 113-8519, Japan

Corresponding author: Toshiyuki Yamamoto, MD, PhD., E-mail: toyamade@fmu.ac.jp

INTRODUCTION

Eccrine syringofibroadenoma (ESFA) is a rare tumor originated from eccrine ductal portion [1,2]. Clinically, ESFA presents a solitary papule, nodule or plaque to multiple lesions. Cases with multiple papular or macular lesions have been often associated with hereditary ectodermal dysplasia [3]. Recent findings have classified ESFA into several groups including, solitary ESFA, multiple ESFA with hydrotic ectodermal dysplasia, multiple ESFA without associated cutaneous findings, nonfamilial unilateral linear ESFA, and reactive ESFA associated with inflammatory or neoplastic dermatoses [4]. We report a case of solitary ESFA which histologically showed various degrees for sweat gland differentiation.

CASE REPORT

A 46-year-old Japanese woman presented with a solitary, 2.3×2.0 cm tumor involving the dorsum of her left foot over 10 years previously. The tumor was erosive, red-colored, and elastic hard (Fig. 1). She, as well as family members, did not have histories of ectodermal dysplasia. Laboratory examination denied diabetes mellitus. Histological examination of the totally resected tumor showed the anastomosing strands of cuboidal cells extending from the epidermis to the upper dermis, with a number of well-defined ducts suggesting eccrine ductal origin. In addition, there were papillary tubular adenoma-like ductal structures lined by a few rows of epithelial cells with papillary projections into the lumen surrounded by fibrous stroma in the mid-dermis. It is of note that various histologic features showing different differentiation were seen in a single lesion of eccrine syringofibroadenoma.

Key words: Eccrine syringofibroadenoma; Papillary tubular adenoma; Foot

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labeled secondary antibody, and finally avidin-biotin-peroxidase complex. Sections were developed with 3,3-diaminobenzidine solution, dehydrated, cleared and mounted. Negative controls were prepared with omitting of primary antibodies and by the substitution by isotype-matched control IgG. CEA was detected on the tumor cells of ductal wall (Fig. 3a). S-100 was detected on the cuboidal cells, whereas negative in the ductal structure of ESFA (Fig. 3b). The neoplastic cells were negative with monoclonal antibodies to desmin and vimentin.

**DISCUSSION**

ESFA have been reported to occur in association with inflammatory disorders such as bullous pemphigoid [5], erosive palmoplantar lichen planus [6], lepromatous leprosy [7], and diabetes mellitus [8,9]. In addition, reactive ESFA has also been reported to occur subsequently on the hyperkeratotic palms and soles [10-15]. Our patient had histories of neither those systemic diseases nor ectodermal dysplasia, but the nodule existed on the dorsum of the foot for long time, suggesting that the tumor was reactive to mechanical stimuli rather than the true neoplasm. In our case, it is of interest that papillary tubular adenoma-like differentiation was partially noted as well as typical ESFA. Papillary tubular adenoma was first described by Abenoza and Ackerman that includes both papillary eccrine adenoma and tubular apocrine adenoma [16]. The histological features are characterized by a relatively well-circumscribed dermal tumor containing numerous tubular and cystic structures with intradermal papillary projections surrounded by fibrous stroma. Thus far, more than 50 cases of ESFA have been reported, however, to our knowledge, ESFA showing papillary tubular adenoma-like lesions has not been reported. Although ESFA is an eccrine tumor, the presence of papillary projections and decapitation-like findings seen in our case favor apocrine lesions. Ishiko et al. [17] also reported the close association of both eccrine and apocrine tumor in a single tumor. On the other hand, two histopathological variants were noted in separate portion of ESFA occurred in one patient [18]. Moreover, syringoma-like dilated sweat duct was seen in the mid-dermis in our case. Sweat gland proliferation has been previously reported in a variety of inflammatory skin diseases, as well as benign and malignant neoplasms [19]. Hara et al. [20] also reported syringoma-like structures intimately associated with epithelial cords in ESFA. Thus, syringoid ductal

![Figure 1: Clinical appearance of the granulomatous tumor on the dorsa of the left foot.](image1)

![Figure 2: Histological view. (a) Lower magnification of tumors. The tumors are consisted of anastomosing epithelial cord strands. Syringoma-like dilated, eccrine duct proliferation is seen in the mid-dermis. (b) Well-defined ductal structures consisted of a few layers. (c) Proliferation of ductal structures lined by basaloïd, elongated tumor cells surrounded by fibrous stroma. (d) Higher magnification. Elongated tumor cells are prominent into the lumen showing decapitation secretion.](image2)

![Figure 3: (a) Immunoreactive cells for CEA are seen in the ductal cells. (b) Immunoreactive cells for S-100 are seen in the epithelial cells.](image3)
proliferation seems not to be rare. In this patient, ESFA occurred on the dorsum of the foot which is susceptible for chronic, recurrent stimuli. Since ESFA is suggested to occur reactively in response to mechanical stimuli, we speculate that various differentiation was also induced during repair process.

Consent

The examination of the patient was conducted according to the Declaration of Helsinki principles.

REFERENCES


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