

A Zosteriform Network of Eccrine Spiradenoma

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Eccrine spiradenoma is clinically characterized by a solitary, movable, tender nodule occurring most frequently on the ventral surface of the upper body. A striking symptom is paroxysmal pain. Occasionally multiple lesions may be present and may occur in a linear or zosteriform pattern.

A 48-year-old female was seen with painful or tender, normal skin colored, firm nodules distributed in a zosteriform pattern on the right thigh for 10 years, which had been slowly growing. The histopathological features were quite a characteristic of eccrine spiradenoma.

We report a rare case of zosteriform network of eccrine spiradenoma occurred on the right thigh. (*Ann Dermatol* 4:(2) 139-142, 1992)

Key Words: Eccrine spiradenoma, Zosteriform pattern

Eccrine spiradenoma is a well defined benign adnexal neoplasm that was first characterized as an entity in 1956 by Kersting and Helwig¹. A typical eccrine spiradenoma appears as a solitary painful, normal skin colored or blue nodule. Rarely a multiple type forming a zosteriform network or linear arrangement has been reported²⁻¹¹.

The present report is illustrative of a eccrine spiradenoma that was especially baffling for the clinician. It appeared as a mysterious painful and tender, unilateral band of nodules distributed on the right thigh.

REPORT OF A CASE

A 48-year-old woman presented at the clinic with five painful and tender nodules distributed on the right thigh for 10 years. The lesion had slowly grown to its present size without prior bleeding, ulceration or crust. No family members had similar lesion.

Examination of the right thigh revealed five normal skin to brown colored, rice grain to chestnut

in size firm grouped nodules observed unilaterally in a zosteriform pattern on the right thigh (Fig. 1). The lesions were raised above the skin surface and ranging from 0.3-2.5cm in diameter. Most of the lesions were flesh colored, however one lesion was dark brown color.

Complete blood cell count, routine urinalysis and liver function test findings were within normal limits. Chest x-ray revealed non-contributory.

Microscopic examination of biopsy specimen revealed typical spiradenoma. The excisional biopsy was done from the lesion of right thigh. In hematoxylin-eosin stain, the tumor consists of sharply demarcated, encapsulated lobules and it was located in the dermis without connection to the epidermis (Fig. 2). The epithelial cells within the tumor lobules were found to be arranged in intertwining cords (Fig. 3). Two types of epithelial cells were noted, intensely staining cells were located at the periphery of the cellular aggregates and pale staining cells were located in the center of aggregates (Fig. 4). In PAS stain, the lumen contained small amounts of granular, eosinophilic material that was PAS positive and diastase-resistant (Fig. 5).

On the basis of clinical and histopathological data, a diagnosis of eccrine spiradenoma was made.

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Fig. 1. Several normal skin to brown colored firm grouped nodules observed unilaterally in a zosteriform pattern on the right thigh.



Fig. 2. The tumor consist of sharply demarcated, encapsulated lobules and it was located in the dermis without connection to the epidermis (H&E stain, $\times 40$).

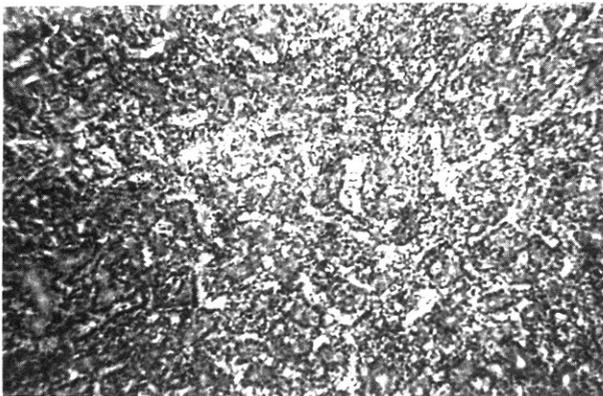


Fig. 3. The epithelial cells within the tumor lobules were found to be arranged in interwining cords (H&E stain, $\times 100$).

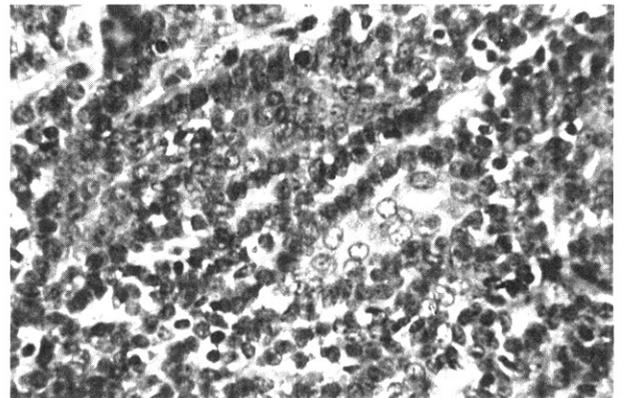


Fig. 4. Two types of epithelial cells were noted, intensely staining cells were located at the periphery of the cellular aggregates and pale staining cells were located in the center of aggregates (H&E stain, $\times 400$).

DISCUSSION

Eccrine spiradenoma is a small, discrete, solitary, well defined nodule, freely movable with the skin or in the immediately subjacent tissue. The tumor is soft to moderately firm in consistency, and the overlying skin frequently has blue or normal skin color. Pain is the most frequent and striking symptom of eccrine spiradenoma. The pain occurred in paroxysms and was never continuous.

Ninety-one percent of the tumors are painful and/or tender. Eccrine spiradenoma is situated mainly on the upper part of the body, usually on the ventral surfaces. The male is involved two times more frequently than the female. The age of onset was between 15 and 35 years of age. Even though most of the eccrine spiradenoma reported are benign, there are reports of the malignant eccrine spiradenoma and malignant transforma-

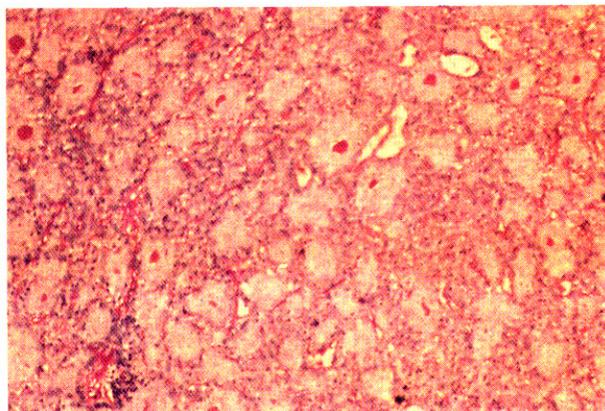


Fig. 5. The lumen contained small amounts of granular, eosinophilic material that was PAS positive and diastase-resistant (PAS stain, $\times 100$).

tion of long-standing¹²⁻¹⁵, apparently benign tumors is well documented. The vast majority of eccrine spiradenoma were solitary lesions. Rarely, a multiple type of eccrine spiradenoma can occur in localized groups^{3, 9, 10, 16}, linear array^{3, 5, 17}, and even zosteriform patterns^{2, 7, 11}. Case of multifocal tumors reported in the literature are summarized in Table 1.

The present case is unusual in that the tumors are not only multifocal occurring in a zosteriform pattern, but also located on the right thigh.

The histological features of eccrine spiradenoma are quite characteristic^{1, 2}. The tumor consists of multiple sharply demarcated lobules. Two types of cells within the lobules and arranged in intertwining bands. One cell with small, dark nuclei is generally located at the periphery of the bands and cells with large, pale nuclei are located in the

center of the bands and around small lumina or ductlike structures. The lumina frequently contain small amounts of a granular, eosinophilic material that is PAS-positive and diastase-resistant¹⁶⁻²⁰.

In our patient, the biopsy specimen of the tumor showed two types of cells and characteristic arrangement of eccrine spiradenoma and PAS-positive.

The histogenesis of the eccrine spiradenoma has caused much controversy. Since Kersting and Helwig¹ had described that the tumor had an eccrine origin, the presence of myoepithelial cells was suggested by Stegmaier and Kersting⁷. However, this was not supported by Ziprkowski and Krakowski⁸, and Munger et al¹⁹ who agreed with the eccrine origin of the tumor.

Subsequently, Hashimoto et al^{16, 20, 21} demonstrated the presence of myoepithelial cells and phosphorylase which is the most characteristic eccrine enzyme. Winkelmann and Wolf²² have reported that eccrine spiradenoma histochemically resembled trichoepithelioma and other basal cell hamartomas. Castro and Winkelmann²³ supported the view that the spiradenoma is a tumor of basal cell differentiation rather than sweat gland origin. More recently, Magrin et al²⁴ described multiple spiradenomas in association with trichoepithelioma and cylindroma in three members of one family.

In conclusion, eccrine spiradenoma is an adnexal tumor of the skin that usually occurs as a solitary lesion and mainly on the upper part of the body, usually on the ventral surfaces. We report

Table 1. Clinical features of patients with multiple eccrine spiradenoma

Case	Year	Authors	Sex/Age	No. of Lesion	Location	Reference
1	1956	Kersting & Helwig	F/-	2	Forehead/Chest	1
2	-	-	F/-	2	Forehead/Back	7
3	1957	Stagmair & Kersting	F/32	4-5	Lt antecubital fossa	8
4	1961	Ziprowski et al	M/18	18	Lt upper extremity	4
5	1961	Berghorn et al	M/43	12	Rt scapula	6
6	1965	Nodl	M/25	200	Trunk/Rt arm	19
7	1966	Hashimoto et al	F/15	6	Rt forearm	5
8	1971	Bazex	F/16	multiple	Post Trunk	16
9	1980	Wood & Shelly	F/41	multiple	Lt neck	9
10	1986	Revis & Chyu	M/47	9	Scalp	10
11	1986	Mi Hung Cho et al	M/47	10	Lt Trunk	25
12	1992	Present case	F/48	5	Rt Thigh	

an unusual case of multiple eccrine spiradenoma occurring in a zosteriform pattern on the right thigh.

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