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# Multiple eccrine spiradenomas in a zosteriform pattern

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## Abstract

Eccrine spiradenoma (ES) typically presents as a solitary tender lesion. Multiple ES is a rare variant of ES and can present in a segmental, linear, blaschkoid, or zosteriform pattern. The etiology of multiple ES is unknown, but several theories have been suggested including a multipotent stem cell origin. We report the case of a 30-year-old woman with multiple painful ES in a zosteriform pattern on the mid-back and abdomen. Skin biopsy of a representative lesion demonstrated a circumscribed tumor nodule encapsulated by a fibrous capsule with diffuse dense basophilic proliferation located in the dermis. The lesions were then excised on two separate sessions without recurrence.

**Keywords:** Eccrine spiradenoma; zosteriform; zosteriform tumors

## Introduction

Eccrine spiradenoma (ES) is a benign, dermal tumor originating from eccrine sweat glands. It typically presents as a solitary painful lesion, but rarely presents as multiple ES. Multiple ES is estimated to comprise less than 2% of cases of eccrine spiradenoma and has increased incidence in females. This diagnosis is associated with an increased risk of malignant transformation. Herein we report a patient with multiple painful ES distributed in a zosteriform pattern.

## Case Synopsis

A 30-year-old woman presented for evaluation of

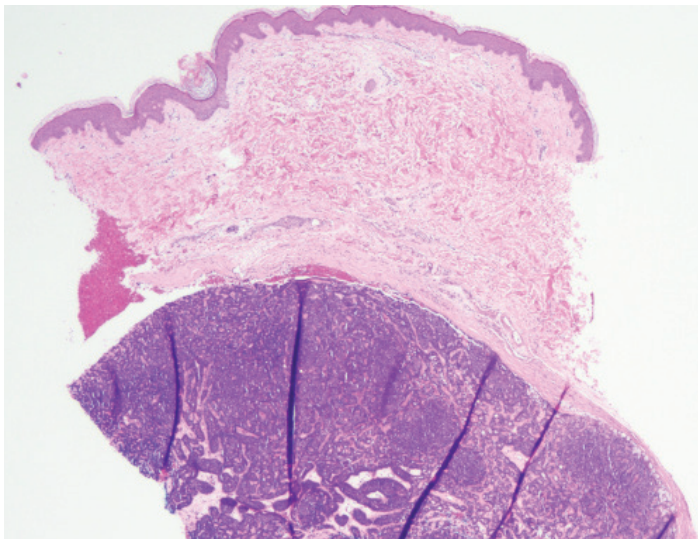
multiple tender nodules localized to the right mid-back and abdomen, which gradually increased in size over the past ten years. She reported the lesions were tender to palpation. The only other comorbidity she presented with was diabetes, managed with metformin. She denied any history of shingles. Review of systems was negative for fever, chills, pain, or weight loss. No significant family history was reported. Physical examination revealed multiple violaceous-pink nodules coalescing into larger plaques on the mid-back extending to the right mid-abdomen in a dermatomal distribution (**Figure 1**).



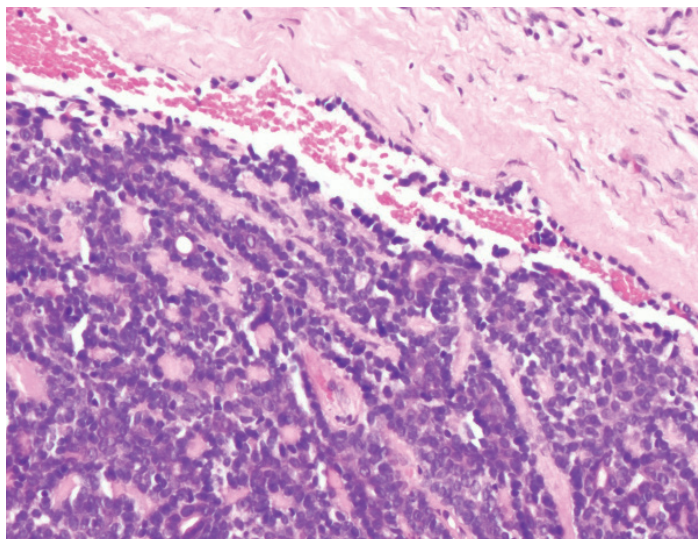
**Figure 1.** Multiple violaceous-pink nodules coalescing into larger plaques on the mid-back extending to the right mid-abdomen in a dermatomal distribution.

A punch biopsy was obtained from a violaceous-pink nodule on the right lower abdomen. Histopathology demonstrated a well-circumscribed nodule of basophilic cells located intradermally with a dense connective tissue capsule surrounding the tumor

(**Figure 2**). On higher magnification, a dense infiltration of basophilic epithelial cells, arranged in trabecular pattern, with blood vessels interspersed throughout the stroma could be observed (**Figure 3**). The nodules consisted of larger basaloid cells with pale nuclei within the center and small cells with hyperchromatic nuclei at the periphery (**Figure 3**). These histologic features supported the diagnosis of eccrine spiradenoma. Given the extent of the lesions, surgical excision was performed in 2 separate stages. There was no complication or recurrence.



**Figure 2.** A well-circumscribed, basophilic dermal nodule with mixed epithelioid and smaller lymphoid cells with dense hyalinized stroma. (H&E, 2x).



**Figure 3.** A mixed population of basophilic epithelioid and smaller hyperchromatic lymphoid cells arranged in rosettes or trabeculae with dense hyalinized stroma. The basaloid cells are large with pale nuclei admixed with smaller hyperchromatic lymphoid cells. (H&E, 20x).

## Case Discussion

Eccrine spiradenomas present as tender, slow-growing, pink-to-violaceous intradermal nodules measuring up to a couple centimeters. A study of 134 ES cases revealed 79% of ES cases were located ventrally; 76% of the ES cases were located on the upper body [1]. ES typically are small solitary lesions but rarely can present as multiple ES and in patterns such as multifocal, linear, zosteriform, or blaschkoid [2-4]. Multiple ES comprised less than 2% of ES cases studied by Kersting et al. [1]. A recent literature review on multiple ES cases showed a female to male ratio of 3:1 in 22 cases with an age of onset ranging from 0-50 years old [2]. ES lesions that are multiple in number have a greater risk for transforming into malignant lesions compared to those that are solitary ES [5].

The etiology of multiple ES is unknown but it has been suggested to arise during embryogenesis from multipotent stem cells of the folliculosebaceous-apocrine unit. These subsequently go on to develop nodules [6]. Trauma has been considered as another possible predisposing factor [7]. Multiple ES with a zosteriform pattern has been suggested to be an organic hamartoma owing to its relationship with other appendageal tumors, such as trichoepithelioma, hidradenoma, and cylindroma [8, 9]. Wright et al. [8] reported multiple ES and multiple cylindroma in three generations of one family, which supports this association. In addition, this study proposed that ES is transmitted in an autosomal dominant manner [8].

Owing to the rare occurrence of multiple eccrine spiradenoma, few studies on its histopathologic features have been conducted. Hashimoto et al. [10] conducted a histochemical, electron microscopic, and light microscopic study on multiple ES and found that the tumor contained two types of cells: the outer basal cells and the inner cells. There were several tumor islands, which had individual fibrous or hyaline capsules. Some of the inner cells expressed secretory cell differentiation [10]. The tumor had glandular and tubular structures, with the tubular structures resembling intradermal eccrine sweat ducts. The tumor contained succinic hydrogenase, branching enzyme, and amylophosphorylase, which support eccrine differentiation [10].

Pain was the most frequent symptom found in the study of 134 ES cases with 91% experiencing pain at the ES lesions and 9% remaining asymptomatic throughout their course [1]. However, another study found pain or tenderness in only 23% of the 49 ES cases [11]. If pain is present, then the differential diagnosis of ES includes the well-known "LEND-AN-EGG" mnemonic, leiomyoma, eccrine spiradenoma, neuroma, dermatofibroma, angioliipoma, neurilemmoma, endometrioma, glomus tumor, and granular cell tumor [1, 12]. If the lesion has no associated pain, then additional diagnoses to be considered include: cylindroma, lipoma, poroma, and trichoepithelioma. For a definitive diagnosis of eccrine spiradenoma, a skin biopsy should be performed. If the histology shows both cylindroma and eccrine spiradenoma features, Brooke-Spiegler syndrome should be considered as a possible diagnosis [13].

Surgical excision is considered the treatment of choice for treating eccrine spiradenoma. There are no specific recommendations regarding margins for standard excision and the recurrence rate is unknown [14]. Other successful reported treatments include CO2 laser ablation, Mohs micrographic surgery, and radiotherapy [2, 14].

## Conclusion

Eccrine spiradenoma is a dermal tumor of eccrine sweat gland origin that may present as multiple ES having a distinct pattern such as zosteriform. There are various theories regarding its etiology. Due to its potential malignant transformation, a skin biopsy should be obtained to diagnose multiple ES.

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