Eccrine Spiradenoma in a Zosteriform Distribution: Presentation of a Case

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To the Editor:

Eccrine spiradenoma is a rare benign adnexal tumor of the eccrine sweat glands; very rarely it can undergo malignant change.1 It usually presents as a solitary nodule and, less commonly, as multiple lesions with a zosteriform distribution.1,2 The clinical diagnosis is often confused with neuromas, leiomyomas, neurilemmoma, neurofibroma, leiomyosarcoma, endometrioma, hidradenocarcinoma of the sweat glands, gliom tumors, lipoma, angiolipoma, dermatofibroma, hemangioma, angioleiomyoma, cavernous hemangioma, or lymphangioma.2,3

There has been a significant increase in the number of cases reported in the literature in recent years,2-5 leading to the suggestion that this diagnosis is suspected more often; this will also lead to greater knowledge of its clinical features.

The patient was a 17-year-old woman with no past history of interest until 7 years earlier, when she started to develop tender, slightly bluish and mildly erythematous nodular lesions, that were round and had very clear margins. They were soft, with a smooth surface, and were about the size of a lentil (3-5 mm). The lesions were in a linear distribution that started in the left popliteal fossa and extended along the dorsal aspect of the thigh up to the inferior part of the ipsilateral gluteal region; 8 lesions were found on dermatologic examination (Figure 1). During the 7-year course of the disorder, the patient had been seen in various hospitals, though the diagnosis had not been reached. The patient therefore came to our hospital where, from a clinical point of view, we considered the following diagnoses: neuromas, neurilemmoma, and neurofibroma. Surgical excision of a nodule was performed for histological study. The histological diagnosis was benign eccrine spiradenoma (Figures 2 and 3); other disorders that could have caused clinical diagnostic confusion were excluded. All the lesions were removed surgically.

Figure 1. Linear topography of the lesions.
Figure 2. Histology (hematoxylin-eosin, low magnification). Tumor separated from the epidermis by a band of normal collagen.
In 1896 Unna coined the term spiradenoma to describe a benign adenoma of the sweat gland arising from the secretory coil of the gland, in contrast to the syringoadenoma, which arises from the ducts.3-5 In 1956, Kersting and Helwig described this condition, referring to it as a rare benign tumor that develops from the secretory and ductal parts of the sweat gland.3-5 In our patient, the condition started when she was 10 years old, some years earlier than those reported as most common in the literature reviewed.3,4 The literature suggests that this disorder usually presents in adolescents and young adults between 15 and 35 years of age, although congenital, familial, and multiple cases have been reported, and also cases related to other benign tumors of sweat glands with autosomal dominant transmission.3-5 This is a rare disease, and much more so in elderly patients. However, in 1998, Senol et al reported the case of a 60-year-old man with a superficial ulcer; the main symptom was spontaneous bleeding of the lesion rather than pain, and the diagnosis was confirmed by histological study.4,5 Although the disease affects both sexes, it is more common in women, with a ratio of 2 to 1.5

In the present case the nodules were situated on the posterior aspect of the left thigh. Other authors indicate that the lesions usually present on the anterior aspect of the trunk or upper limbs, but cases have been reported on the head (scalp, nasal cavity, and pinna of the ear); it is rare to find them on the palms of the hands, in the axillas, or on the areola, perineum, or genitalia, as these are areas in which there is a predominance of apocrine glands.1 There are no reports of this tumor affecting a mucocutaneous junction or the nail bed.3-5

Our patient presented multiple, subcutaneous, nodular lesions that were very small (3-5 mm) and tender, and showed a linear (zosteriform) distribution. A similar condition to that of this patient has been reported as a rare form,1-4 though it was described at the same site (dorsal aspect of the thigh). These tumors may be associated with cylindromas and trichoepitheliomas. One case has been reported with multiple lesions with a linear, nevoid distribution affecting the right side of the body from the face to the pubis.1,3,4 The clinical course is chronic and, very rarely, malignant transformation can occur, with the possibility of developing metastases in regional lymph nodes, bone, lung, and brain, and it can be fatal.2,4 In the case presented here, there was no association with any other skin tumor nor was there malignant transformation of the lesions.

There has been a significant increase in the number of cases reported in the literature in recent years.1-3 We would therefore suggest that this diagnosis be kept more in mind. We consider it important to report the present case due to the low frequency of presentation of linear (zosteriform) eccrine spiradenoma and, in particular, because it has not been reported on the dorsal aspect of the thigh.

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Conflicts of Interest
The authors declare no conflicts of interest.

References