INTRODUCTION

Eccrine spiradenoma (ES), an uncommon benign adnexal tumor of sweat gland origin, occurs mainly in young adults, equally in both sexes.\(^1\) It is characterized by a solitary, painful, deep-seated nodule on the trunk or proximal limbs.\(^1\) Multiple ES in a zosteriform distribution is extremely rare and only few cases have been reported in literature.\(^2\) Here, we describe a patient with multiple ES in a zosteriform distribution involving three dermatomes.

CASE REPORT

A 35-year-old woman presented with multiple, elevated lesions on the lower part of the face, trunk, and left upper extremity for the last 4 years. The lesions were insidious in onset but had become painful and increased in size for the last 2 years. Pain was intermittent and dull aching in nature.

On examination, multiple, erythematous to dusky, firm, tender nodules of different sizes, varying from 0.5 to 4 cm in diameter, were arranged in a linear pattern starting from the midline and extending along the chin and left submandibular region as well as over the front and back of the left side of chest and over the flexor aspect of the left arm and forearm [Figure 1a-c]. The nodules were along the distribution of mandibular branch of trigeminal nerve (V3), C3, and T1 dermatomes, respectively. Palms, soles, mucous membranes, hair, and nails were normal. Differential diagnoses considered were ES, neurofibroma, leiomyoma cutis, dermatofibroma, and angiolipoma.

Routine hematological and biochemical investigations were normal. Mantoux test was negative. Punch biopsy of a 1 cm sized nodule on hematoxylin and eosin stain revealed variable sized lobular aggregates surrounded by condensed hyaline connective tissue in the dermis [Figure 2a]. At higher magnification, the lobules were observed to be containing ductal structures with pale cells arranged around the lumina and dark cells in the periphery [Figure 2b].

ABSTRACT

Eccrine spiradenoma is an uncommon benign tumor of the sweat glands, most frequently characterized by a solitary, painful, deep-seated nodule. A case of multiple spiradenomas in a zosteriform distribution involving three dermatomes is described with its clinico-pathological features.

Key words: Spiradenoma, three, zosteriform
A final diagnosis of zosteriform spiradenoma was made based on clinical and characteristic histopathologic findings. Surgical excision of the tumors was advised, but the patient refused.

**DISCUSSION**

ES is a benign tumor arising from the transitional area between the secretory portion and the coiled duct of the eccrine sweat gland.\(^4\)

Clinically, it presents as a solitary, rounded, bluish, firm, and painful dermal nodule about 3–50 mm in diameter.\(^5\)

The usual site is front of trunk and proximal limbs.\(^1\)

In our patient, tumors were distributed along specific dermatomes. Zosteriform and linear distribution of ESs is extremely rare where the tumors have been proposed to develop from an organic hamartomatous process frequently associated with different appendageal components.\(^4,6\) Shelley and Wood\(^2\) suggested neural origin for zosteriform spiradenomas.

Histologically, the tumor commonly consists of several sharply demarcated lobules in the dermis without connections to the epidermis. The lobules may display a fibrous capsule and are often deeply basophilic due to dense packing of nuclei.\(^7\) Two types of epithelial cells are present in the tumor lobules; those with small, dark nuclei arranged in the periphery and those with large, pale nuclei arranged in the center of the cellular aggregates.\(^1,7\)

Markedly atypical cells with high mitotic figures, necrosis, loss of dual cell population, and lobular architecture indicate malignant transformation.\(^8\) No evidence of a malignant transformation was noted in our case.

Spontaneous episodes of pain may occur in spiradenoma which has been attributed to contraction of myoepithelial cells, but electron microscopy shows the absence of myoepithelial cells in the tumor.\(^3\) The exact explanation for pain is thus lacking.

Malignant transformation, though rare, can occur in long-standing lesions.\(^3\) Complete surgical excision is the treatment of choice.\(^1\) Recurrence may occur if lesions are partially excised.\(^1\) If there are multiple tumors, surgical excision may not be practicable.\(^9\) Radiotherapy and CO\(_2\) laser are the alternative treatment modalities available in such cases.\(^4\)

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**Conflicts of interest**

There are no conflicts of interest.

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