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FACIAL ECCRINE SPIRADENOMA MASQUERADING AS BENIGN APPENDAGEAL TUMORS-AN UNUSUAL PRESENTATION.

Dermatology

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ABSTRACT

Eccrine Spiradenoma lesions are usually solitary and, the common sites include front of the trunk and proximal limbs. This study reports an atypical presentation of eccrine spiradenoma in which multiple lesions are distributed over the face, being discrete and non-tender unlike the classical tender, and solitary lesion or zosteriform pattern of lesions seen on the trunk. Review of literature did not reveal any such case report.

KEYWORDS

Eccrine Spiradenoma, multiple lesions, face, ablative fractional CO2 laser.

INTRODUCTION:

Eccrine Spiradenoma (ES) is a benign tumor of the sweat gland lineage of unknown etiology. It is relatively uncommon, occurring mainly in young adults. Rarely, it can be familial or may occur as a part of the Brooke-Spiegler syndrome.[1] The lesion is usually a painful, solitary, bluish, firm, rounded 3–50 mm dermal nodule occurring on trunk and proximal limbs.[1] Lesions have also been described following Blaschko lines or in a linear or zosteriform distribution.[1] We are reporting a 46-year-old female patient presenting with asymptomatic multiple eccrine spiradenomas at an unusual site and multiplicity of lesions.

CASE REPORT:

A 46-year-old female patient came to the DVL OPD with complaints of asymptomatic, multiple, solid, raised lesions over the face since five years. Patient observed a gradual onset of few solid raised lesions of varying sizes ranging from 0.5 cm to 1.5 cm over her forehead, which later progressed to involve the midline of face, cheeks and chin. Lesions were associated with itching and increase in size on exposure to sunlight and sweat. There was no history of secondary changes like ulceration or blister formation, rapid increase in the size of the lesion or other lesions present anywhere else on the body. There was no history of loss of sensations or hypopigmented patches or peripheral nerve thickenings. Drug history was not significant. She did not have any systemic complaints. Family history for similar lesions was negative. Systemic examination was unremarkable.

On cutaneous examination: multiple, discrete, erythematous to skin colored, shiny, firm, dome-shaped, non-tender, papulo-nodular lesions of 0.5 cm to 1.5 cm size were distributed over the forehead and midline of the face. Few scattered lesions were seen over the cheeks and the submandibular area. The skin was non-pinchable.

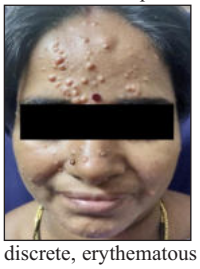

Figure 1A. Multiple, discrete, erythematous to skin coloured, shiny, dome-shaped, papulo-nodular lesions of 0.5-1.5 cm size distributed over the forehead, nose, B/L cheeks and chin regions.

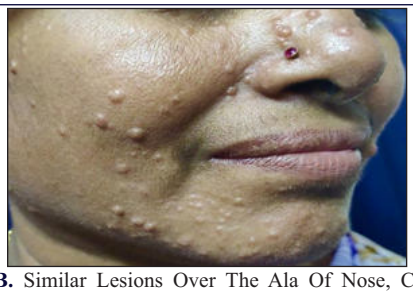

Figure 1B. Similar Lesions Over The Ala Of Nose, Cheek And Mandibular Region.

Hair, nails and mucosa did not show any abnormalities. Differential diagnoses considered were Trichoepithelioma, Cylindroma, Sebaceous hyperplasia, Benign Appendageal Tumor. Routine biochemical investigations were within normal limits, and a 3mm punch biopsy was done. Histopathology revealed a tumor proper, having rounded nests with two types of epithelial cells arranged in intertwining cords in the dermis and subcutis. One type of cells were dark basaloid with small dark nuclei and scanty cytoplasm; another type of cells were pale with vesicular nuclei and moderate cytoplasm. Hyaline globules were present within the tumor nests, and the intervening stroma was edematous. There was no connection between the tumor nests and the surface epithelium, and there was no evidence of atypia. All these histological features were suggestive of Eccrine Spiradenoma without any malignant transformation.

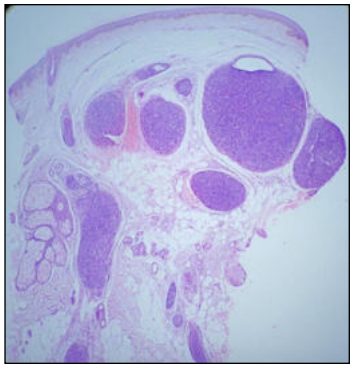

Figure 2A. Scanner View.

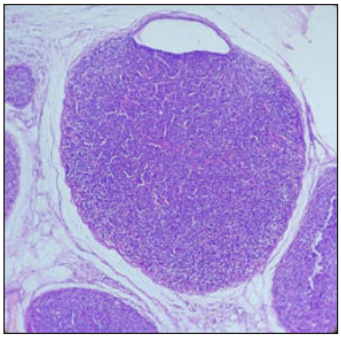

Figure 2B. Tumour proper with rounded nests in the dermis and subcutis on H&E, Low power (10x).

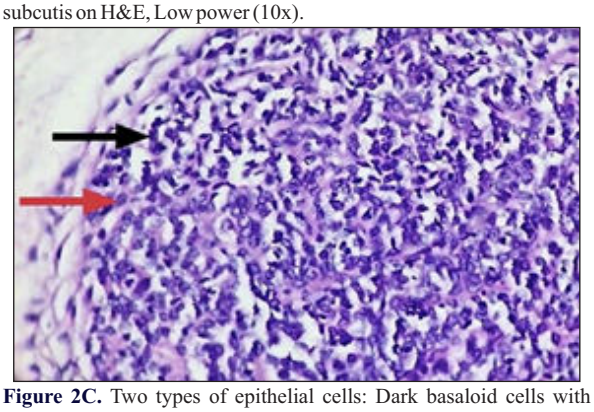

Figure 2C. Two types of epithelial cells: Dark basaloid cells with small dark nuclei, scanty cytoplasm (black arrow) and Pale cells with vesicular nuclei, moderate cytoplasm (red arrow), arranged in intertwining cords in the dermis and subcutis on H&E, High power (40x).

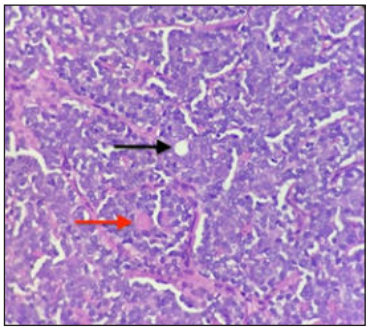

Figure 2D. Hyaline globules (red arrow) and ductal structures (black arrow) were present within the tumor nests.

Since lesions were multiple and located on the face, surgical excision of each discrete lesion was not feasible. Instead, Fractional CO_2 laser ablation in repeated sessions was done but the results were not that satisfactory. Patient was referred to a plastic surgeon for further treatment options.

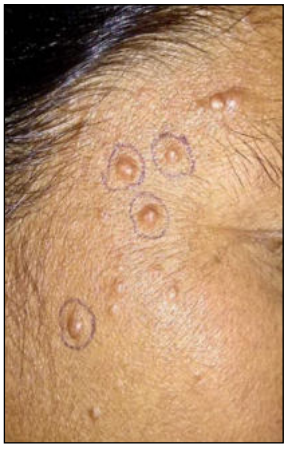

Figure 3A. Lesions before treatment with CO_2 laser.

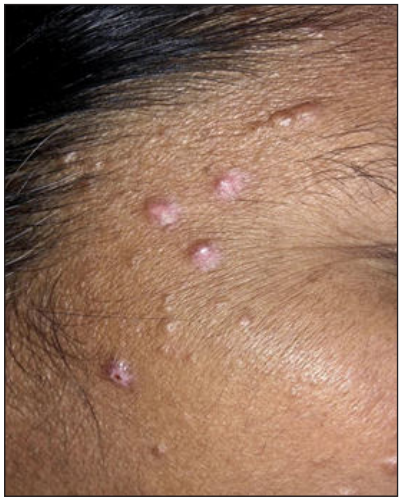

Figure 3B. Improvement with resolving lesions seen two weeks after CO_2 laser treatment.

DISCUSSION:

Most presentations of ES are solitary and do not show any sex predilection. However, multiple ES affects women predominantly, with a female/male ratio of 3:1.[2] Multiple ES can be described either as multifocal or localized with a linear zosteriform distribution and blaschkoid or nevoid pattern.[2] Saha M. et al. presented a case showing multiple eccrine spiradenomas in a zosteriform distribution involving three dermatomes including the face in a 33-year-old female patient.[3] Ren F. et al. also presented a case of multiple segmental eccrine spiradenomas in a zosteriform pattern in a 32 year old female patient.[2] The proposed mechanism of multifocal ES is that during embryogenesis, an abnormal clone of multipotent stem cells of the folliculo-sebaceous-apocrine unit arises, which produces abnormal cells resulting in nodule formation and a possible malignant transformation.[2] The spontaneous episodes of pain that may occur in ES have been attributed to myoepithelial cells' contraction, but myopeithelial cells are absent on electron microscopy. Hence, the exact explanation for pain is unclear.[3] In our case, a 46-year-old female patient, multifocal eccrine spiradenomas were present on the face and did not have a zosteriform or blaschkoid pattern, and the lesions were not associated with pain or tenderness. Histopathology played a cornerstone in unveiling the diagnosis. Malignant transformation is more common in longstanding tumours and in multiple ES. Recurrence may occur if lesions are partially excised; therefore, complete surgical excision is the treatment of choice.[1] Surgical excision is not practical when there are multiple lesions, but alternate modalities available for such cases include radiotherapy and CO_2 laser.[3] In our case, Fractional CO_2 laser ablation in repeated sessions was tried with unsatisfactory outcome so patient was referred to a plastic surgeon.

CONCLUSIONS:

Occurrence of multiple Eccrine Spiradenoma lesions on face is quite unusual and unreported in the world literature, thus bringing out the uniqueness of this case.

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