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Original Research Paper

Dermatology

NEVUS UNIUS LATERIS – A CASE SERIES

Dr Sreesha C* — Resident, Department of Dermatology, Venereology & Leprosy, Chettinad Hospital & Research Institute, Kelambakkam, 603103. *Corresponding Author

Dr Sankeerthana M P — Resident, Department of Dermatology, Venereology & Leprosy, Chettinad Hospital & Research Institute, Kelambakkam, 603103.

Dr Jayakar Thomas — Emeritus Professor, Department of Dermatology, Venereology & Leprosy, Chettinad Hospital & Research Institute, Kelambakkam, 603103.

ABSTRACT Verrucous epidermal nevi (VEN) are a common type of keratinocyte hamartomas which are often linear in distribution. Nevus unius lateris (NUL) is a rare systematised variant of VEN with unilateral involvement. It is often associated with disorders in the nervous system, musculoskeletal system, visual and auditory disturbances. The sites commonly affected are trunk and limbs, but it can also affect the head and neck region or the oral mucosa rarely.[1][2]

KEYWORDS : Nevus unius lateris, verrucous epidermal nevus, Cutaneous mosaicism.

INTRODUCTION

Verrucous epidermal nevus is a common clinical finding and originates from the embryonic ectoderm, but Nevus unius lateris, which is a systematised variant of VEN, is not very common. The etiopathogenesis is not very well known.[1][2] It is characterised clinically by confluent verrucous, papillomatous plaques that align with the Blaschko lines and are distributed linearly and involves only one half of the body. The onset tends to be at birth or early in life.[3] Musculoskeletal, neurological, visual and auditory abnormalities are often associated with this disorder.[1] It is important to identify the comorbidities associated with NUL for therapeutic and morbidity purposes.

Case Report 1

A 17-year-old female patient presented to the Dermatology outpatient department with hyperpigmented, verrucous plaques in the right hemibody at the level of the axilla, trunk, abdomen and arm [Figure 1]. These lesions were present since birth and increased in size progressively. Genitals and mucous membranes were spared. Normal psychomotor development was noted in all stages of life. The patient had unremarkable personal and maternal history. The lesions were asymptomatic for 15 years, however as the lesions grew in size, they also became pedunculated and painful. There were no other abnormalities noted at the time of presentation. Biopsy was taken from the lesion on left arm and histopathological examination showed psoriasiform hyperplasia of epidermis and parakeratosis. Electro-desiccation was done to improve cosmetic complaints but patient did not come back for follow up.

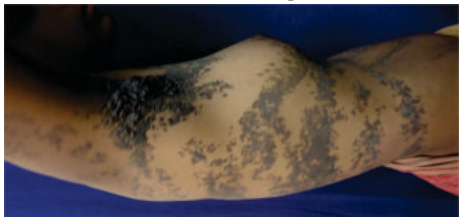

Figure 1: Hyperpigmented verrucous plaques on the right side of the trunk, abdomen and on right arm. Note that the lesions over the right axilla are more verrucous.

Case Report 2

A 25-year-old male patient presented to our Dermatology OPD with asymptomatic, confluent hyperpigmented verrucous plaques on left side of posterior trunk and nape of the neck [Figure 3] which started in infancy and progressed gradually since then to current size and configuration. There was no involvement of oral mucosa. No history of any delay in developmental milestones noted. No history of similar complaints in the family members was noted. Biopsy was taken from the upper back lesion and the findings were suggestive of nevus unius lateris. Electro surgery was advised to the patient as a modality of treatment but the patient did not consent for it.

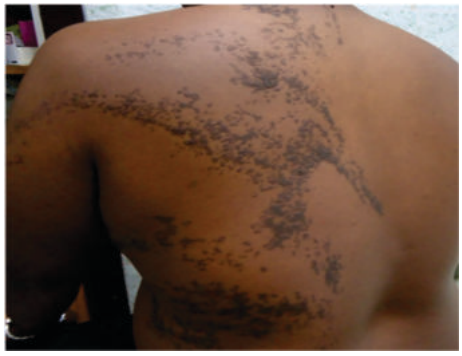

Figure 2: Confluent hyperpigmented verrucous plaques on left side of posterior trunk and nape of the neck.

Case Report 3

An 18-year-old female patient presented with linear, hyperpigmented, verrucous plaques on the antero-lateral aspect of left thigh along the lines of Blaschko [Figure 3]. The lesions started shortly after birth and progressed gradually in size and became more prominent and pigmented. There were no symptoms associated with the lesions. Patient's personal and maternal history were unremarkable. No history of any developmental delay. No other comorbidities were noted. Patient refused for a biopsy to be taken and was lost to follow up.

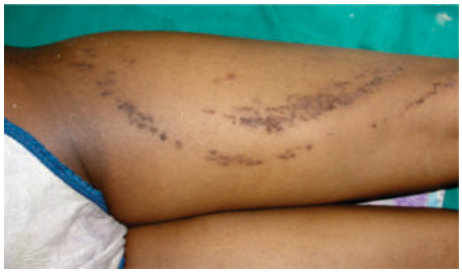

Figure 3: Lesions on the anterior aspect of the left thigh along the lines of Blaschko.

Case Report 4

A 20-year-old male patient presented to the outpatient department with multiple coalesced hyperkeratotic plaques on the lateral aspect of right leg [Figure 4]. On palpation, the lesions were rubbery and did not bleed on touch. No history of seizures, mental retardation or skeletal abnormality noted. A punch biopsy was taken from this site and the results showed hyperplastic epidermis along with thickening and elongation of rete ridges and normal adnexal structures [Figure 5 & 6]. The patient was advised to have three to four cryotherapy sessions spaced one month apart. Right now, the patient is in between the second and third sessions.

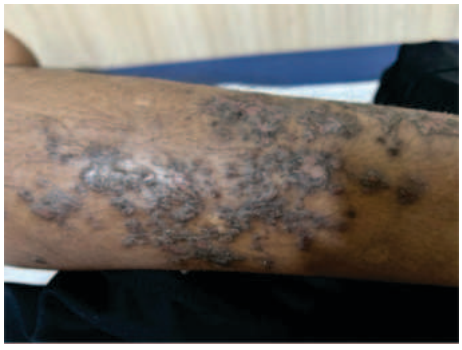

Figure 4: Hyperkeratotic plaques seen on the lateral aspect of the right leg.

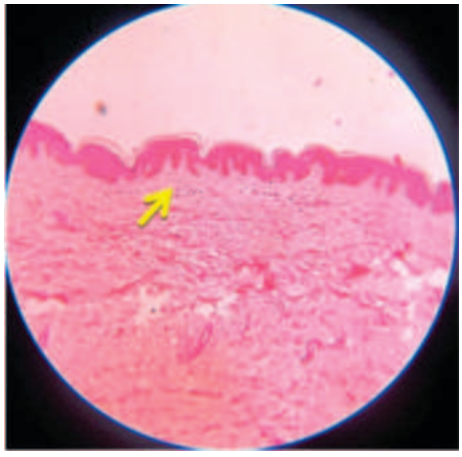

Figure 5: Under low power, the epidermis shows papillary epidermal hyperplasia with thickening and elongation of rete ridges leading to psoriasiform appearance. There is inflammatory infiltrate in the dermis as indicated by the arrow.

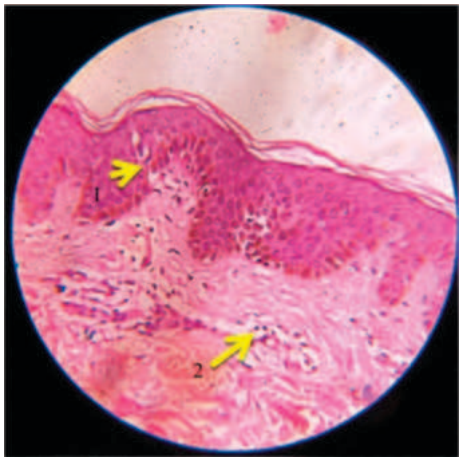

Figure 6: Under high power, the epidermis shows thickening of the rete ridges along with spongiosis. The papillary dermis shows perivascular lymphocytic infiltrate.

DISCUSSION

VEN is a form of cutaneous mosaicism which is a result of de-novo postzygotic mutations. Nevus unius lateris is a type of VEN which is confined to only one side of the body. Although the disease's etiopathogenesis is not yet fully known, if the nevus follows Blaschko's lines, it is classified as mosaicism.[1] It is characterised by confluent, hyperpigmented verrucous plaques limited to a hemibody. The sites commonly involved are the trunk and extremities.[2] Rarely, we see the head and neck involvement. Clinical diagnosis is considered gold standard more often than not, but in some cases a biopsy may be required for confirmation. The histopathological findings include hyperkeratosis, acanthosis, papillomatosis and elongation of rete ridges which corresponds clinically to the raised lesions with a verrucous surface.[3] The lesions remain asymptomatic in most cases but the risk of traumatic detachment and subsequent erosions and secondary bacterial infections is high if the lesions become pedunculated and/or appear in flexors. The management of large lesions is very difficult. It is possible to employ cryotherapy, electrofulgration, photodynamic therapy, CO2 laser, surgical methods, calcipotriol, and systemic & local retinoids. Recurrences are common and the outcomes are not constant. In severe situations, the outcomes are anti-aesthetic. Scars usually replace the verrucous lesions.[1][4][5][6]

CONCLUSION

This condition is rarely described in the literature previously. It is often associated with various systemic abnormalities such as seizures, stunting of growth, neoplasms of the brain as well as disturbances in vision and hearing, and hence prompt diagnosis and evaluation is extremely important. These cases have been presented here for its rarity and for further research to be done in this direction to improve the quality of lives of the affected patients.

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