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Case Report

Dermatology Section

Verrucous Haemangioma with Histoid Leprosy: A Case Report

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ABSTRACT

Verrucous Haemangioma (VH) is a rare, congenital vascular malformation of cutaneous and subcutaneous tissue. It usually presents as macular lesion at birth which progresses with age, and may become bluish papules, nodules, and warty plaques. Linear presentation of this disease is even rarer. Here the present case report presents a case of a 30-year-old male patient with multiple raised erythematous hyperkeratotic papules and plaques over right forearm since birth which was arranged in a linear pattern.The patient also had asymptomatic papules on right thigh and lower back region, associated with sensory loss and peripheral nerve thickening. Dermoscopy of forearm showed reddish-blue lacunae, alveolar pattern, and whitish veils. Colour Doppler showed intradermal dilated venous channels with thrombosis which was suggestive of haemangioma. Histopathological examination confirmed the diagnosis of linear VH on forearm and slit skin Ziehl-Neelsen staining was suggestive of histoid leprosy. Linear VH is a rare congenital venous malformation, progresses proportionately with age. Dermoscopy can help in early diagnosis of vascular lesions. Complete surgical excision is the treatment of choice.

Keywords: Dermoscopy, Forearm plaques, Surgical excision, Venous malformation

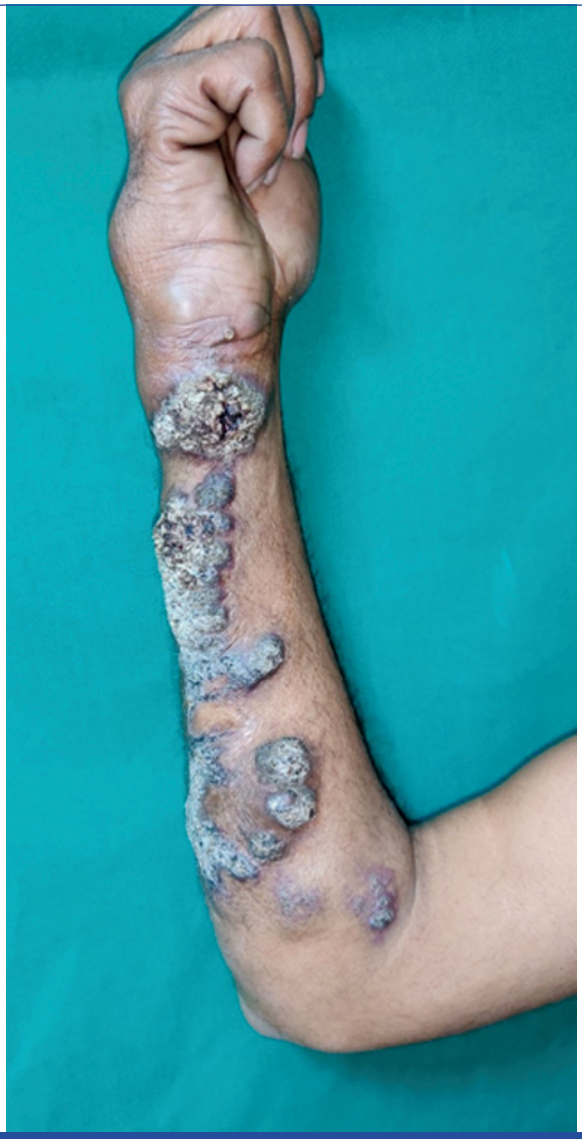
CASE REPORT

A 30-year-old male presented to the dermatology and venereology outpatient department with multiple raised reddish-purple lesions on the inner aspect of his right forearm since birth. These lesions were initially flat and small but gradually thickened and enlarged over time. within a span of 6-7 years, they progressed to involve the entire forearm. The patient had a history of surgical excision of a similar lesion 20 years ago, but it reappeared within one year and became more prominent and thicker than before. There were no significant history of trauma or systemic symptoms.

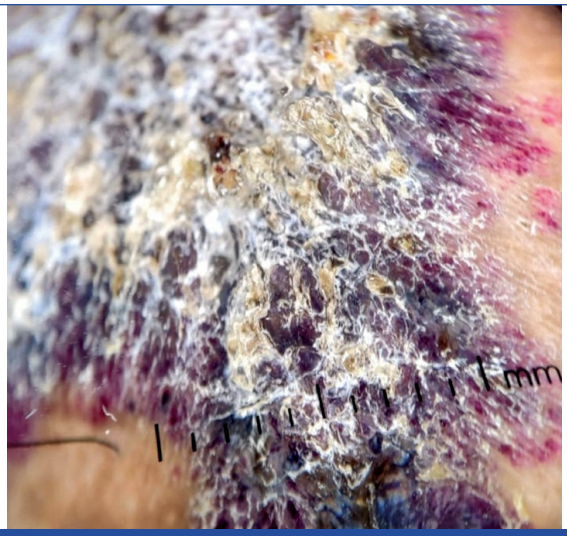
On examination, multiple erythematous to violaceous hyperkeratotic papules, nodules, and plaques of varying sizes (ranging from 1×1 cm to 5×7 cm) were observed in a linear pattern on the medial aspect of the right forearm. Central fissuring and necrotic areas were present, along with surrounding peripheral erythema and a few satellite lesions of similar morphology [Table/Fig-1]. Dermatoscopic examination revealed an alveolar pattern, prominent hyperkeratotic lesions, and whitish veils over them, indicating mature lesions [Table/Fig-2]. The periphery of the lesion showed reddish-blue lacunae characteristic of vascular lesions [Table/Fig-3]. The patient also had a few reddish, translucent, asymptomatic papules of size 0.3×0.5 cm with normal skin underneath on the lower back and anterior aspect of the right thigh [Table/Fig-4]. Sensory loss over the medial aspect of the right hand and dorsal aspect of the right foot, as well as peripheral nerve thickening involving the right ulnar nerve and right common peroneal nerve, were noted.

Colour Doppler sonography of the verrucous lesion revealed intradermal dilated venous channels with thrombosis and intimal calcification in some lesions, suggesting haemangioma. Sonography also detected a potential lesion on the dorsal aspect of the hand that was not clinically appreciable. Based on these features, a differential diagnosis of linear VH, angiokeratoma circumscriptum, and leprosy was considered. A skin biopsy was performed on the forearm lesion using a 4 mm punch, and a slit skin smear examination using Ziehl-Neelsen staining was planned for the papule on the lower back.

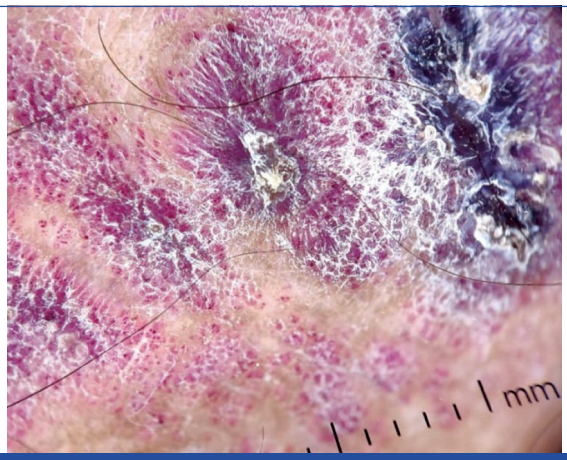
Histopathological examination {Haematoxylin and Eosin (H&E)} of the biopsy specimen from the forearm showed epidermal compact



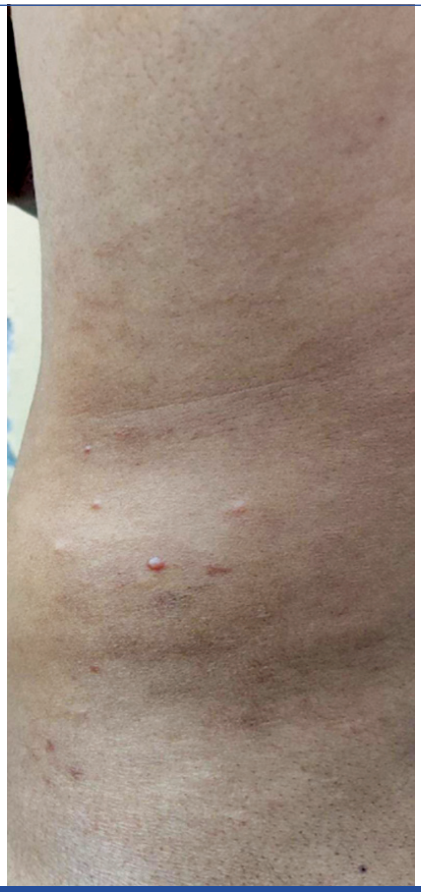
[Table/Fig-1]: Erythematous to violaceous hyperkeratotic papules and plaques with central fissuring in a linear pattern over the medial aspect of right forearm.



[Table/Fig-2]: Dermoscopic examination showing alveolar pattern along with very prominent hyperkeratotic areas and whitish veils.

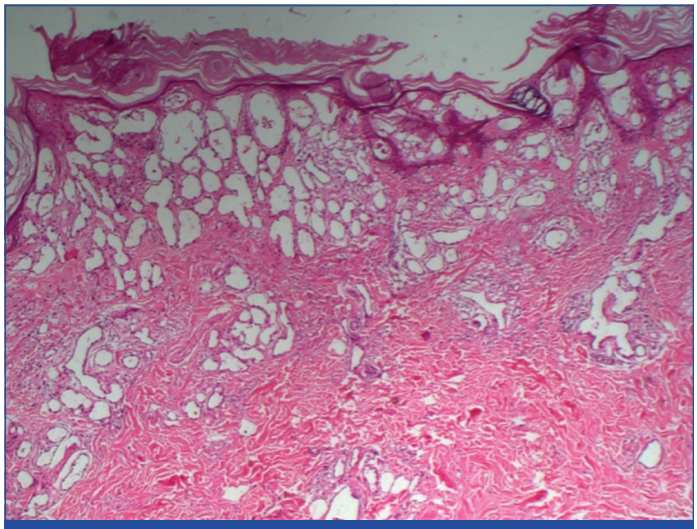


[Table/Fig-3]: Dermoscopic examination of peripheral area of the lesion showing reddish-blue lacunae, characteristic of vascular lesions.

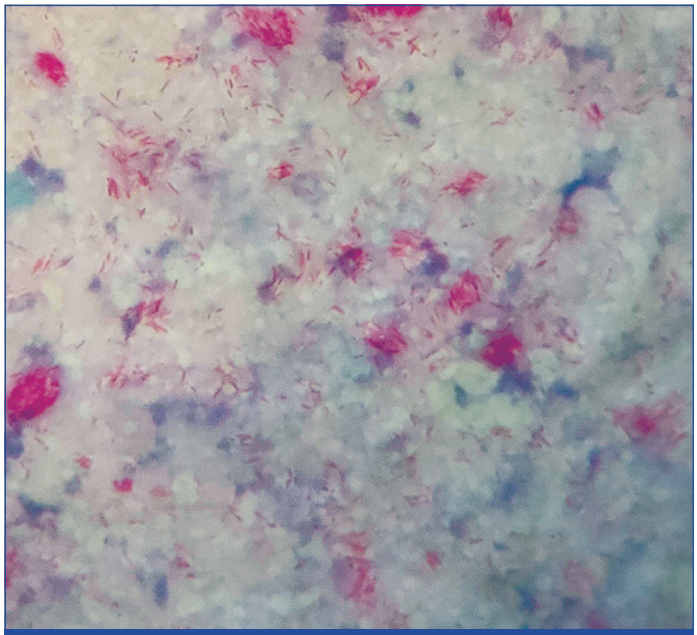


[Table/Fig-4]: Reddish translucent papule over lower back.

ortho-hyperkeratosis with slight acanthosis. The papillary dermis, reticular dermis, and subcutaneous tissue were infiltrated by clusters of dilated thin-walled capillaries [Table/Fig-5]. Ziehl-Neelsen staining smear of the lesion on the back revealed multiple solid-stained elongated acid-fast bacilli, suggestive of histoid leprosy [Table/Fig-6].



[Table/Fig-5]: Photomicrograph showing epidermal ortho-hyperkeratosis with clusters of variable-sized dilated, thin-walled capillaries in the thickened papillary dermis, reticular dermis and subcutaneous tissue. (H&E, x40).



[Table/Fig-6]: Multiple solid-stained elongated acid-fast bacilli (Ziehl-Neelsen stain, x100).

Therefore, a diagnosis of linear VH with histoid leprosy was made. The patient was managed for histoid leprosy with multibacillary multiple drug therapy and was referred to a plastic surgeon for further management of VH. Although complete surgical excision of VH was advised, the patient refused and was prescribed clobetasol propionate 0.05% with salicylic acid 6% ointment to be applied twice daily. After two months of treatment, there was no significant response in the haemangioma lesion, and the patient was subsequently lost to follow-up.

DISCUSSION

Halter K introduced the term “VH” in 1937, which is a misnomer as it is an uncommon vascular malformation [1]. VHs are uncommon congenital vascular malformations that were first described by Imperial R and Helwing EB in 1967 [2]. In 2018, the International Society for the Study of Vascular Anomalies (ISSVA) classified vascular lesions into three main classes: vascular tumors, vascular

malformations, and unclassified anomalies, with VH belonging to the category of verrucous venous malformation [3]. The causative gene associated with verrucous venous malformation is MAPK3K3 [3]. Due to the rarity of this disease, there is no published data regarding its incidence or prevalence.

The VH typically presents at birth or in early childhood. The initial lesions are non keratotic, soft, bluish-red macules that gradually increase in size and number with age, leading to the formation of thickened papules and plaques with surrounding erythema. VH is most commonly found as unilateral, localised, and scattered lesions on the lower limbs (approximately 95%) [4]. However, it may also occur in uncommon sites such as the abdomen, upper limb, and penis [5]. Rarely, VH may present in a serpiginous, reticular, or linear arrangement [6]. The linear arrangement of these lesions represents genetic mosaicism. Complications such as recurrent episodes of bleeding and infections result in the characteristic bluish-black verrucous hyperkeratotic surface [7]. Important differential diagnoses include infantile hemangioma and salmon patch in the early stage, and angiokeratoma circumscriptum and Verrucous Epidermal Nevus (VEN) in the matured stage.

Dermoscopic features of VH include an alveolar appearance with numerous bluish small ovals to polygonal elements, as well as veils with different shades of blue, including light blue [8]. In the present case, dermoscopy revealed more prominent hyperkeratosis along with bluish lacunae indicating the underlying dilated vascular channels, consistent with VH. Histopathologically, VH is characterised by irregular acanthosis and hyperkeratosis in the epidermis, along with abnormal dilated vascular channels located in the dermis and subcutaneous tissue. It is essential to differentiate VH from angiokeratoma, where dilated vascular channels are limited to the papillary dermis. Immunohistochemistry can be used to confirm the diagnosis, with clusters of venules showing positivity for Glucose Transporter 1 (GLUT1) [9]. In the patient, histopathology was suggestive of VH, but immunostaining was not performed due to financial constraints.

Complete surgical excision is the treatment of choice for VH. Incomplete excision may lead to persistence or recurrence of the lesion. The recurrence rate of VH is 33%, especially when the lesion is larger than 2 cm, primarily due to deeper vascular infiltration [2]. Other therapeutic options include topical corticosteroid with salicylic acid combination ointment, cryosurgery, electrocautery, and various laser treatments, such as pulsed dye laser, CO_2 laser, and Neodymium-doped Yttrium Aluminum Garnet (Nd:YAG), for lesions smaller than 2 cm, although their efficacy is limited [10].

A literature review of cases with linear VH involving the upper limb is summarised in [Table/Fig-7] [11-14].

Name of author/Year of study	Age (years)/ Gender	Location	Morphology	Dermoscopy
Wentscher U and Happle R (2000) [11]	16/Male	Left upper limb	Multiple hyperkeratotic plaques of different sizes	NA
Nupur P et al., (2014) [12]	7/Male	Right upper limb	Multiple hyperkeratotic erythematous plaques	NA
Nargis T et al., (2017) [13]	7/Male	Left upper limb	Multiple hyperkeratotic erythematous plaques	NA
Al-Furaih I et al., (2020) [14]	13/Male	Right upper limb	Multiple hyperkeratotic plaques of different sizes	NA
Present case (2022)	30/Male	Right upper limb	Multiple erythematous to violaceous hyperkeratotic papules and plaques	Showed reddish-blue lacunae, alveolar pattern and whitish veils

[Table/Fig-7]: Literature review of cases with linear Verrucous Haemangioma (VH) over upper limb [11-14].
NA: Not applicable

CONCLUSION(S)

Linear VH is a rare disease, with very few cases reported from India. Lesions involving the upper limb are even rarer. In the present case, authors performed dermoscopic evaluation, which can be a helpful tool in diagnosing such a rare vascular malformation.

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