Multiple Verrucous Hemangiomas: A Case Report with New Therapeutic Insight

Abstract
Verrucous hemangioma is an uncommon congenital vascular malformation, which may clinically masquerade angiokeratoma, lymphangioma circumscriptum, or malignant melanoma. Differentiation is essential owing to varied therapeutic and prognostic implications. We present a rare case of multiple verrucous hemangiomas in a teenage girl who presented with multiple warty lesions over the dorsal aspect of the left foot since birth. Magnetic resonance imaging (MRI) scan was suggestive of a vascular malformation, and skin biopsy showed ectatic blood vessels extending from the papillary dermis into the subcutaneous tissue, diagnostic of verrucous hemangioma. A combination of 0.05% halobetasol propionate with 3% salicylic acid ointment was advised. This therapeutic intervention resulted in significant resolution of the warty lesions over a period of 2 months, following which surgical excision was performed. The implication is that we can use a combination of super potent topical steroid with salicylic acid as an adjunct to surgical resection.

Keywords: Hemangioma, topical steroid, verrucous

Introduction
Verrucous hemangioma is an uncommon, localized, congenital vascular malformation which presents as a bluish red lesion in the early stages and eventually acquires a warty surface as a protective phenomenon.[1] Clinically, lesions may simulate angiokeratoma, lymphangioma circumscriptum, verrucous epidermal nevus, or even malignant melanoma.[2] Although thorough clinicoradiological examination helps in narrowing the differential diagnosis, histomorphological assessment of this masquerader is imperative owing to therapeutic and prognostic implications.

We describe a rare case of multiple verrucous hemangiomas in a teenage girl with numerous warty lesions over the left foot. Our case highlights the clinical, radiological, histopathological, and immunohistochemical features of verrucous hemangioma along with utility of topical steroids as a therapeutic intervention.

Case Report
A 15-year-old female presented to the outpatient dermatology department with chief complaints of brownish painful warty lesions over the dorsum of the left foot since birth. Initially, the lesions were red, soft, and gradually increased in size and number up to the age of 5 years to become warty, brownish black, hard, and tender. History of bleeding usually following minor trauma was noted. General physical examination was within normal limits. Cutaneous examination revealed multiple hyperpigmented, hyperkeratotic plaques consisting of closely-set warty papules varying from 2 cm × 2.5 cm to 3 cm × 3.5 cm involving the dorsum of the left foot and lateral malleolus [Figure 1]. Surface of the lesion was firm and verrucous with no ulceration, bleeding, or atrophy. The lesion was noncompressible and diascopy was negative. No other hair, nail, or mucosal abnormality was noted. There was no palpable regional lymphadenopathy. Radiographic films of the left foot showed normal findings. Color Doppler revealed ill-defined heteroechoic soft tissue densities at the anterior, superior, and lateral aspect of the left foot showing few echogenic foci and slow venous flow. Magnetic resonance imaging (MRI) scan showed mild delayed enhancing lesion in the skin and subcutaneous plane of the left foot. A probable diagnosis of vascular malformation, capillary venous type,
was given and skin biopsy was performed. Hematoxylin and eosin (H and E) sections from skin biopsy showed hyperkeratosis, parakeratosis, acanthosis, and papillomatosis of the epidermis. Underlying papillary dermis showed multiple thin-walled and ectatic blood filled spaces with mild perivascular mononuclear cell inflammatory infiltrate [Figure 2a]. Similar vascular channels were also seen in the mid dermis and subcutaneous tissue [Figure 2b]. These channels were lined by flattened endothelial cells. On immunohistochemistry (IHC), the endothelial cells showed positivity for cluster of differentiation 34 (CD 34) [Figure 2c], CD 31, and Wilm’s tumor 1 (WT-1) [Figure 2d]. Based on the immunomorphological features a final diagnosis of verrucous hemangioma of the left foot was rendered. Patient was started on a combination of topical 0.05% halobetasol propionate with 3% salicylic acid ointment twice daily. The lesions reduced in size and became less warty over a period of 2 months [Figure 3]. Patient was then referred to plastic surgery for excision. Postoperative period was uneventful with no recurrence after 1-year follow-up.

Discussion

Verrucous hemangioma is an uncommon, congenital vascular malformation first denominated by Imperial and Helwig in 1967.[3] These are most frequently located on the lower extremities with rare cases reported on the head, trunk, upper limbs, and glans penis. Although lesions are usually solitary and localized, a rather uncommon variant of multiple disseminated lesions without systemic involvement has been described.[4] Our case revealed similar multiple verrucous lesions over the dorsal aspect of the left foot. However, there was no disseminated pattern or systemic involvement.

Clinically, lesions of verrucous hemangioma may closely simulate angiokeratoma, and differentiation is essential because incomplete excision of verrucous hemangioma would lead to recurrence of the lesion from the deeper components.[5] MRI is the diagnostic modality of choice because it delineates the dermal and subcutaneous plane of these lesions and guides biopsy.[6] Histopathological examination remains the gold standard for diagnosis with presence of ectatic channels extending into the deeper dermis vis-à-vis angiokeratoma circumscripturn [Table 1]. Immunohistochemical positivity of endothelial cells lining the vascular channels for CD 34 and CD 31 antibodies acts as an adjunct.

The accurate categorization of verrucous hemangioma still remains elusive. To clarify the nosology of this vascular anomaly, Trindade et al. first reported WT-1 positivity in 13 cases of verrucous hemangiomas.[7] Similar immunohistochemical results were obtained in our case. However, the recently identified genetic mutation in the Ang1-TIE2 (angiopoietin 1-tunica internal endothelial cell kinase) pathway confirms that verrucous hemangioma has genetic features consistent with a venous malformation, despite having an immunoprofile similar to vascular neoplasms.[8] This also opens the door for targeted therapy.

Verrucous hemangiomas do not resolve spontaneously and early diagnosis is important for timely surgical excision and better cosmetic result. Superficial ablative procedures such as electrocautery and cryosurgery invariably lead to recurrence of the lesion.[9] Thus, surgery remains the mainstay of treatment. While topical steroids have a
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promising result in most superficial hemangiomas, the therapeutic role of this treatment modality has never been studied in the setting of verrucous hemangiomas. Numerous theories explaining the mechanism of steroids have been published with the concept of vasoconstriction being widely accepted.[10] A combination of halobetasol propionate with salicylic acid ointment used in our case resulted in significant resolution in 2 months following application. Salicylic acid aids in the penetration of the topical steroid in this warty hyperkeratotic lesion owing to its keratolytic action. The implications of this therapy is that we can use a combination of super potent topical steroid with salicylic acid to reduce the size of these lesions, which would aid in the ease of subsequent surgical resection. However, due to paucity of literature, the potential utility and outcome of this therapeutic modality cannot be ascertained.

Conclusion

To conclude, this case was presented for its atypical presentation as multiple warty lesions. Moreover, our aim was to diminish the size of the warty lesions using a combination of halobetasol propionate with salicylic acid, with the intention of reducing the amount of bleeding at surgery and better cosmetic results. Further studies are recommended to study the response of these lesions to this combination therapy.

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Conflicts of interest

There are no conflicts of interest.

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Table 1: Comparison between verrucous hemangioma and angiokeratoma circumscriptum

| Features                  | Verrucous hemangioma                  | Angiokeratoma circumscriptum          |
|---------------------------|---------------------------------------|---------------------------------------|
| Onset                     | Congenital                            | Acquired                              |
| Clinical presentation     | Usually solitary;                     | Punctate lesions varying from 1 mm to 5 mm coalesce to form plagues; |
|                           | Size of the lesion varies from 1 cm to 7 cm; | Bleeding is infrequently present      |
|                           | Oozing and/or bleeding is often present | Multiple thin-walled and ectatic blood vessels are seen are seen limited to the superficial dermis only |
| Histopathological         | Multiple thin-walled and ectatic blood vessels are seen extending from the superficial dermis to deep dermis | Present |
| distinguishing feature    | Present                               | Absent                                |
| WT-1 immunohistochemical  |                                       |                                       |
| expression                |                                       |                                       |
| Treatment                 | Surgical excision                     | Cryotherapy, electrocautery, laser    |
| Recurrence                | Common                                | Rare                                  |

Figure 3: Significant resolution of lesions two months after halobetasol and salicylic acid ointment application