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Linear verrucous hemangioma—a rare case and dermoscopic clues to diagnosis

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ABSTRACT Verrucous hemangioma (VH) is a rare, congenital and localized vascular malformation, which usually presents as warty, bluish, vascular papules, plaques, or nodules, mainly on the lower extremities. Linear presentation of the disease is rare. A deep biopsy is necessary to confirm the clinical diagnosis by histopathological examination, with dermoscopy acting as a useful tool for evaluating the precise vascular structure. Here, we report on a 13-year-old female child with linear VH presenting over her foot since infancy and dermoscopic findings of VH along with the clinical-pathologic features.

Introduction

Verrucous hemangioma is an uncommon congenital vascular malformation described by Imperial and Helwig as a separate entity in 1967 [1]. It usually presents at birth or in early childhood, with the commonest site being the lower extremities. The initial lesion presents as a reddish macular area resembling a vascular stain. Gradually with the growth of child, the lesions increase in size, spread locally and become verrucous. The lesions are usually scattered but linear, serpiginous and reticular patterns can be seen rarely [2]. The linear arrangement of these lesions usually reflects genetic mosaicism or dermatomal distribution [3]. Verrucous hemangioma does not involute spontaneously, rather, incomplete excision can result in regrowth [3]. Clinically, VH is a close mimicker of other vascular lesions like angiokeratoma, infantile hemangioma and venous or lymphatic malformations. Thus, dermoscopic and microscopic evaluation aids in confirming the clinical diagnosis.

Case Report

A 13-year-old female presented with purplish, warty skin lesions over the inner surface of the left foot. Her mother stated that these lesions were noticed in early infancy, and with age they enlarged, increased in number, and became irregular on the surface. There was no history of any trauma or bleeding from these lesions. Cutaneous inspection revealed well-defined erythematous to violaceous plaques and nodules with verrucous surfaces arranged in a linear array over the medial aspect of the left foot that were tender on palpation (Figure 1a, b). No limb length discrepancy was noted. Systemic examination did not reveal any abnormalities, nor did laboratory investigations.

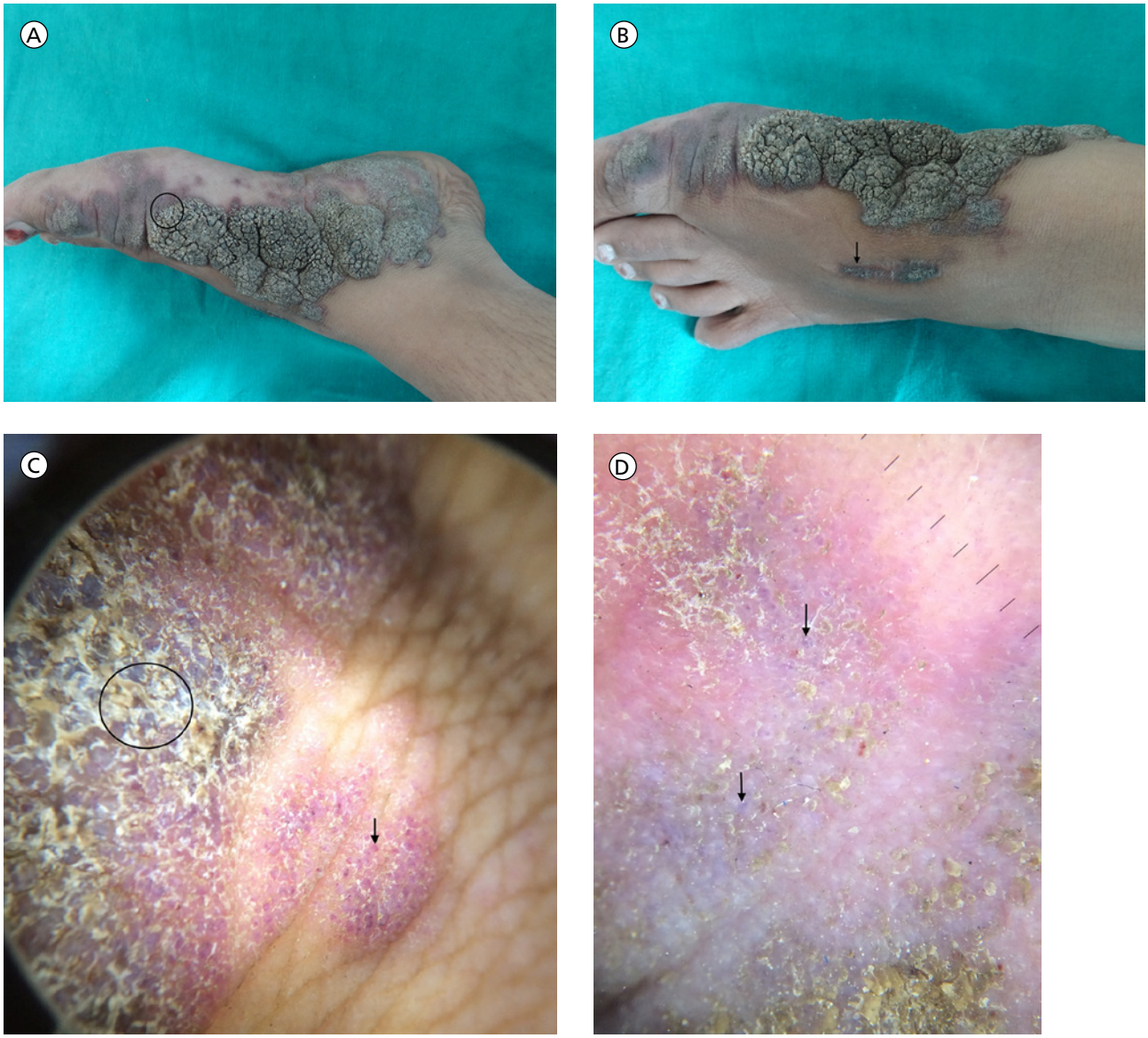

Figure 1. (a) Erythematous to violaceous plaques with verrucous surface (black circle) arranged in a linear array over the medial aspect of the left foot. (b) Satellite plaques (black arrow) arranged linearly over the dorsum of foot. (c) Dermoscopy of verrucous lesions showing the prominent hyperkeratosis over bluish background (black circle) along with the reddish blue lacunae (black arrow) indicating the underlying dilated vascular channels. (d) Peripheral areas of the lesion showing the bluish lacunae (black arrow) characteristic of vascular lesions correlating with the vascular channels seen in histopathology. [Copyright: ©2018 Dhanta et al.]

The dermoscopic features were different depending on the type and site of the lesions. The most striking feature was a prominent bluish background. Hyperkeratosis and bluish lacunae were observed in most of the lesions but they were most prominent in verrucous plaques (Figure 1c). The periphery of the plaque showed well-defined dark blue lacunae characteristic of vascular lesions(Figure 1d).

Doppler sonography of the lower limb (arterial system) showed diffuse irregular echogenicity in the subcutaneous plane of the affected area of the left foot. At some places, minimum vascularity was observed and these sonographic features were suggestive of hemangioma.

A 4 mm punch biopsy was taken for histopathological evaluation. Microscopy revealed elongation of rete ridges, thick parakeratosis and dense infiltration of eosinophils in a parakeratotic epidermis. Small and thin-walled capillaries lined by flattened endothelial cells were seen predominantly in the dermis (papillary and deep dermis) reaching up to the dermosubcutaneous junction. A few capillaries were dilated and filled with fresh fibrin thrombi (Figure 2a-c). These features, thus, confirmed the diagnosis of verrucous hemangioma. The patient was referred to plastic surgery, where serial excisions were planned to remove the whole lesion.

Discussion

In 1937, Halter introduced the term "verrucous haemangioma" which is an uncommon vascular malformation. It was first described as a separate entity and distinguished

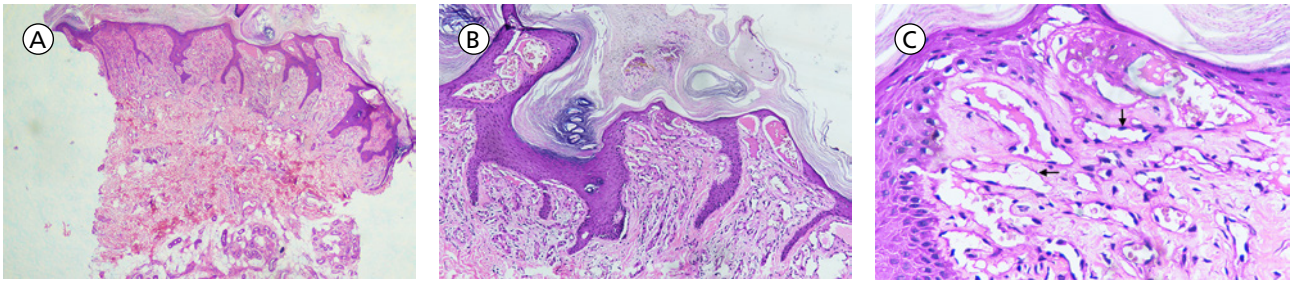

Figure 2. (a) Scanning view showing hyperkeratosis, elongation of rete ridges, and vascular dilatations in the papillary dermis extending to subcutaneous tissue (H&E, 4x). (b) Hyperkeratosis, papillomatosis, acanthosis, and dilated blood vessels in the dermis (H&E, x10). (c) Small and thin-walled dilated capillaries seen in dermis lined by flattened endothelial cells (black arrow) with fibrin thrombi present in some of the vessels (H&E, x40). [Copyright: ©2018 Dhanta et al.]

from angiokeratoma and its other variants by Imperial and Helwig in 1967 [1].

VH usually presents at birth or in early childhood and then gradually progresses in size with age. The initial presentation is a reddish macular area of a vascular anomaly resembling a "port-wine" stain. Recurrent episodes of bleeding and infection result in the characteristic bluish-black color along with the development of a verrucous, hyperkeratotic surface [4]. Localized or scattered lesions on the unilateral lower extremity are the most common presentation. However linear, serpiginous, or reticular pattern can also be seen, though uncommonly [2]. In our literature search, we could find only eight reported cases of linear verrucous hemangioma, making our patient the ninth [1,2,5-10].

Apart from the clinical picture, confirmation of diagnosis is made with histopathology. Characteristic microscopic features of VH are irregular acanthosis and hyperkeratosis in the epidermis. The abnormal proliferating vascular channels are located in the dermis and hypodermis, which differentiates it from angiokeratomas, where the lesion is limited to the papillary dermis. Currently, no specific immunohistochemical marker exists to diagnose VH. In one study, positivity for glucose transporter protein 1 (GLUT1), a determinant expressed by infantile hemangioma, was seen in 7 of 11 VH lesions [11].

The dermoscopic features reported in the literature include an alveolar appearance with numerous small, oval to polygonal elements surrounded by slightly darker pigmentation. Different shades of blue, including light blue, indigo blue, dark bluish black and a bluish white veil have been described [12,13]. Well-defined dark lacunae were seen in the periphery in our case, which is characteristic of vascular lesions and further correlated well with the vascular channels seen on histopathology. Dermoscopy of the late lesions showed more prominent hyperkeratosis along with the bluish lacunae indicating the underlying dilated vascular channels (DermLite II Hybrid M [3Gen, San Juan Capistrano, CA, USA]; 10 × magnification). In our case we could not find a significant alveolar pattern, but all the other features were seen. The highlight of our case was the expected dominant hyperkeratosis seen in the verrucous lesions over a bluish background, which again favored a vascular etiology.

VH in its pre-verrucous state may be indistinguishable from infantile hemangioma, venous or lymphatic malformation and angiokeratoma. Dermoscopy of infantile hemangioma has been reported to exhibit a polymorphous pattern of vascular structures with or without red linear and red dilated vessels [14]. The absence of a bluish component in hemangioma can help distinguish it from VH in early stages. As mentioned by Osio et al. [14], the color of the hemangioma can help classify infantile hemangioma with the superficial type showing a bright reddish color, and the superficial and deep type depicting a dark red color. We would like to believe that a prominent bluish component in VH is seen due to the depth of the vascular involvement. Angiokeratoma has been described as having three patterns in dermoscopy with dark lacunae and whitish veil in all three, peripheral erythema as a second pattern and hemorrhagic crust as a third [15]. Although it is difficult to distinguish between angiokeratoma and VH on the basis of dermoscopy the presence of reddish-blue lacunae without whitish veil, as seen in our case points more towards VH. More studies are needed to differentiate angiokeratoma from VH concretely.

In its mature phase, the clinical and dermoscopic differential diagnoses of VH include pigmented lesions like pigmented basal cell carcinoma, verrucous epidermal nevus and seborroheic keratosis in smaller lesions.

Blue or dark lacunae are rarely seen with dermoscopic examination of non-vascular lesions. Complete absence of a pigment network which is a highly specific dermoscopic feature of melanocytic lesions, helps in differentiating the two. The absence of leaf-like and spoke-wheel pigmentation, arborizing vessels, and erosions separates pigmented basal cell carcinoma from VH. Verrucous epidermal nevus (VEN) shows a large brown circle represented by oval or round structures with a hyperchromic brown edge surrounding a hypochromic area. In VEN dermoscopic pattern is brown in color given its superficial nature and never blue, which helps in differentiating it from VH [16]. Seborrheic keratosis shows milia-like cysts, comedo-like openings, and fissuring

TABLE 1. Dermoscopic findings of verrucous hemangioma and its differential diagnosis

1.	Verrucous Hemangioma	Alveolar appearance with various shadows of bluish small, oval to polygonal elements surrounded by slightly darker pigmentation with well-defined dark lacunae in the periphery. Dominant hyperkeratosis seen in the verrucous lesions [12,13]
2.	Infantile Hemangioma	Polymorphous pattern of vascular structures with or without red linear and red dilated vessels [14]
3.	Angiokeratoma	Dark lacunae and whitish veil, peripheral erythema, and hemorrhagic crust in third pattern [15]
4.	Pigmented Basal Cell Carcinoma	Leaf-like and spoke-wheel pigmentation, arborizing vessels, erosions, blurred lacunae that may look like blue-gray ovoid nests [12]
5.	Verrucous Epidermal Nevus	Large brown circle seen as oval or round structures with a hyperchromic brown edge surrounding a hypochromic area [16]
6.	Seborrheic Keratosis	Milia-like cysts, comedo-like openings, fissures and ridges and sharply demarcated border [17]

without any vascular lacunae, which helps in distinguishing it form VH [17]. The dermoscopic findings of the differential diagnosis of verrucous hemangioma is summarized in Table 1.

Dermoscopy can help play an important role in clinching the diagnosis and aiding in management when clinical findings alone or an inadequate (superficial) biopsy specimen is misleading. The importance of reaching an accurate diagnosis cannot be overemphasized when planning treatment, given that the treatment of choice for VH is complete surgical excision. Incomplete excision leads to persistence, recurrence and continued enlargement of the lesion. Due to the deeper vascular infiltration, the recurrence rate of VH is 33%, especially when the lesions are larger than 2 cm in diameter [3]. Various other options that have been tried with limited results include ultrasound, cryosurgery and electrocautery, especially for smaller lesions [4,18,19]. Recently, a combination of CO2 and dual pulsed dye laser Nd:YAG has been reported to provide satisfactory response in some cases [20].

Conclusion

In this report, we describe clinical, dermoscopic and histopathological features of VH in a 13-year-old girl. We emphasize that VH has distinct dermoscopic features and suggests that dermoscopy can contribute significantly to diagnosing such a rare congenital vascular malformation. The interesting fact about this case is rarity of disease more so than the linear presentation along with the presence of both verrucous and pre-verrucous stages. Further, the dermoscopic pattern in both the types of lesions is another highlight of this case.

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