Non-targetoid Hobnail Hemangioma: Two Case Reports and Literature Review

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ABSTRACT:

Hobnail hemangioma (HH) or targetoid hemosiderotic hemangioma is a benign vascular tumor with classic presentation of pale and ecchymotic halo. However, there are diversity in clinical and dermoscopic manifestations of HH, which can make diagnosis challenging in cases where the characteristic targetoid appearance is absent. This report discussed two cases of non-targetoid HH. A 15-year-old Thai female presented with erythematous plaque on her left knee, where dermoscopic findings include red and dark lacunae with white structures and a reddish homogeneous area, which are indicative of HH. The second case involves a 67-year-old Caucasian male presented with an ulcerated crusted nodule on his upper back. In this case, histopathological examination was necessary to rule out skin cancer and confirm the diagnosis of HH. This study provides an overview of HH and the challenges posed by non-targetoid presentations, emphasizing the importance of clinical and dermoscopic evaluation, as well as histopathological examination in uncertain cases.

Key words: Hobnail hemangioma, Targetoid hemosiderotic hemangioma, Superficial hemosiderotic lymphovascular malformation, Dermoscopy

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Introduction

Hobnail hemangioma (HH), also known as targetoid hemosiderotic hemangioma is a rare benign vascular tumor. A classic presentation of HH is a solitary vascular lesion with a characteristic targetoid appearance, featuring a central brown to violaceous papule surrounded by a pale area and a peripheral ecchymotic ring. However, non-targetoid, indistinctive, and atypical presentation are not uncommon. These variable clinical presentations make diagnosis more challenging. HH may be clinically confused with other benign conditions like as angiokeratoma, dermatofibroma and insect bite reaction, as well as malignant skin lesions such as Kaposi's sarcoma, melanoma, and basal cell carcinoma. Herein, we present two cases of HH with non-targetoid presentations.

Case reports

Case 1

A 15-year-old Thai female presented with asymptomatic dark red rash on her left knee. The rash first appeared at the age of two months and had enlarged following minor trauma 5 months prior. Physical examination revealed a well-circumscribed annular erythematous plaque with violaceous spots on the left knee (Figure 1). Dermoscopic examination showed red and dark lacunae with white structures and reddish homogeneous area (Figure 3). Histopathological examination showed thin-walled dilated vascular spaces lined by bland endothelial cells with few hobnail nuclei in the upper dermis and hemosiderin-laden macrophages scattered throughout the dermis (Figure 2). The diagnosed with HH was made and surgical excision of the lesion was performed.

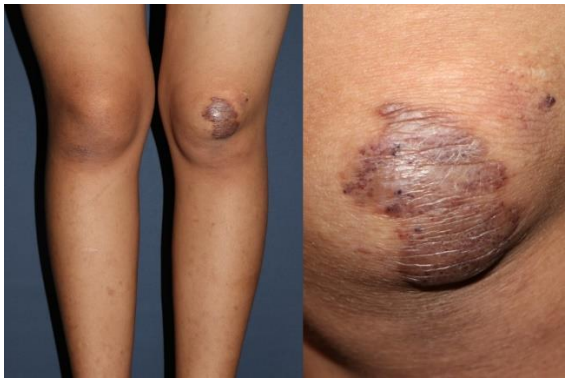

Figure 1 A well-circumscribed annular erythematous plaque with violaceous spots on the left knee

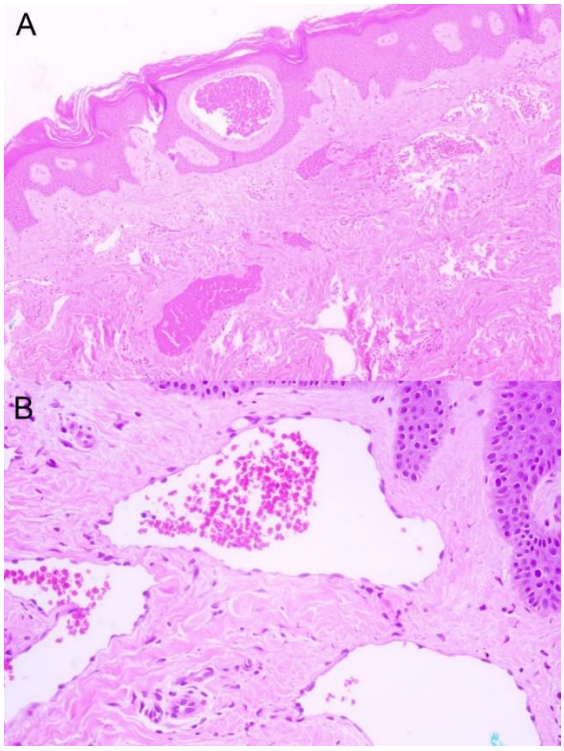

Figure 2 Histologic examination was a HH (hematoxylin-eosin, original magnification X40 in A and X200 in B)

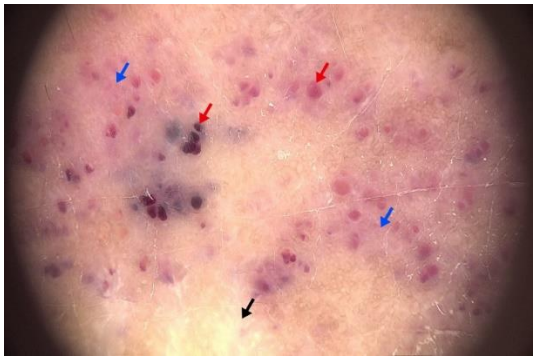

Figure 3 Dermoscopically, we observed red and dark lacunae (red arrows) with white structures (black arrow), and reddish homogeneous areas (blue arrows)

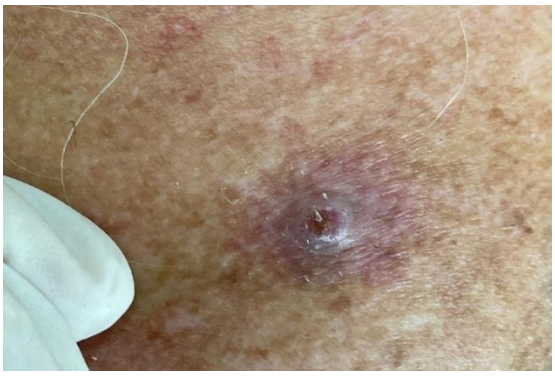

Figure 4 A solitary erythematous nodule with central ulceration and crust on upper back with the background of sun-damaged skin

Case 2

A 67-year-old Caucasian male presented with six-month history of an ulcerated, crusted nodule with surrounding erythema on his upper back (Figure 4). Due to suspicion of skin malignancy, excisional biopsy was performed. Histopathological analysis revealed epidermal crusts with marked spongiosis, vascular proliferation, dilated vascular spaces with thin-walled lining and hobnail endothelial cells in the dermis (Figure 5). Based on histopathologic findings, the diagnosis of HH was established.

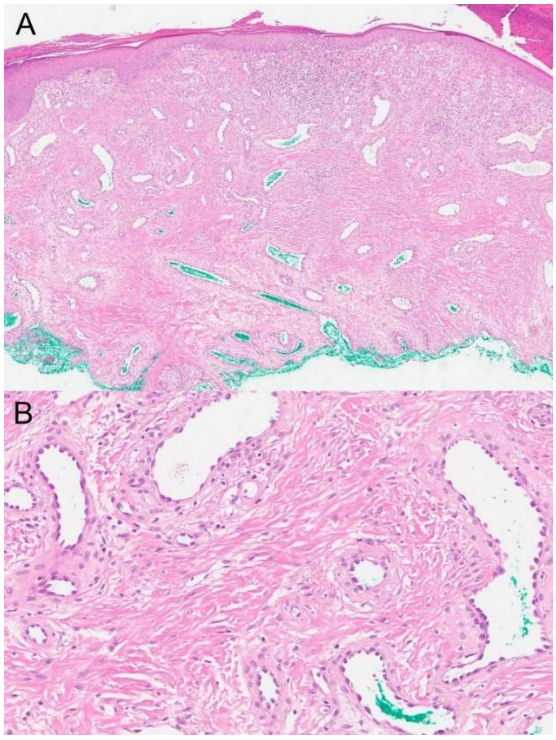

Figure 5 Histologic examination showed HH with epidermal crusts and marked spongiosis (hematoxylin-eosin, original magnification X40 in A and X200 in B)

Discussion

In 1988 Santa Cruz and Aronberg first reported targetoid hemosiderotic hemangioma[1]. The naming emphasizes its characteristic targetoid appearance and deposition of hemosiderin. In 1999, Guillou et al introduced the term hobnail hemangioma (HH) to highlight the distinctive hobnail cytomorphology of the endothelial cell lining the vascular spaces[2]. Thereafter, with the use of immunohistochemical staining, reports unveil a lymphatic origin of the vascular lesion and components of malformation rather than a neoplastic nature[3, 4]. In 2014, Joyce et al proposed renaming it as superficial hemosiderotic lymphovascular malformation[2]. These terms all refer to the same disease entity.

HH typically occurs in children to middle-aged adults without a gender predisposition. It commonly presents as a solitary small papule on the trunk or extremities, but multiple lesions, congenital onset, and a family history have also been reported[5]. The exact etiology of HH remains uncertain. The proposed causation included chronic inflammation, trauma, especially injury to preexisting vascular lesion[2]. Although our case 1 has a clear history of lesion's exacerbation following trauma, the majority of HH cases have negative trauma history. Intermittent changes related to pregnancy and menstruation have been reported, suggesting roles of estrogen and progesterone in pathogenesis of the HH as in many vascular lesions[5, 6].

HH exhibits variable clinical presentations. The classic targetoid appearance is characterized by a brownish to violaceous papule to plaque surrounded by a pale thin area and ecchymotic circular band. However, this characteristic appearance is not always presented as it is a result of hemorrhage and hemosiderin deposition which can spontaneously resolve[6]. With aging, HH can lose its halo and become fibrotic resembling dermatofibroma[7]. Episodic involution and recurrence have also been reported[6]. Clinical variations include non-targetoid lesion, pigmented lesion, skin-colored nodule, dusky red plaque, and other nondistinctive lesions[7]. Due to the dynamic and diverse presentation, clinical differential diagnosis of HH is broad, including benign conditions such as single angiokeratoma, hemangioma, dermatofibroma, nevus, insect bite reaction, along with malignant lesions such as Kaposi's sarcoma, melanoma/amelanotic melanoma, and basal cell carcinoma[8]. Dermoscopy can be a valuable tool in diagnosis of HH. Zaballos et al studied dermoscopic findings in 35 cases of HH. The most common dermoscopic pattern was central red and/or dark lacunae with a peripheral brown homogeneous area (71.4%), followed by reddish or reddish-brown homogeneous area with vascular and white structures (22.8%)[6]. Differential diagnosis to these features includes targetoid hemosiderotic nevi and vascular tumors respectively. Interestingly, Enei et al have reported a case of HH with arborizing vessels, blue-gray oval structure, and multiple brown dots with diffuse brown pigment on dermoscopic analysis, which led to false-positive diagnosis of basal cell carcinoma[9]. Other dermoscopic findings were delicate pigment network, red lacuna alone, skin-colored/yellow/white circular homogeneous area, chrysalis, cerebriform pattern, and gray background[7, 10]. From all reports, the most indicative dermoscopic features of HH might be a lagoon of variable color, regardless of targetoid pattern[7]. However, combination of various colors and/or vascular pattern prompt the suspicion of malignant skin tumors and histopathological examination is essential for a definitive diagnosis[8, 10].

The histopathologic features of HH, like its clinical presentation, have a variable morphology depending upon the stage of biopsied lesion. In early stages, dilated, irregular, thin-walled, vascular channels in the superficial dermis lined with a single layer of hobnail epithelial cells are characteristic findings in the central area. While in deeper and peripheral areas, there are irregular, thin-walled, slit-shaped vascular spaces dissecting between dermal collagen bundles. Hemosiderin deposits, extravasated erythrocytes, and a mononuclear inflammatory infiltrate are commonly found. In later stages, collapsed vascular lumina, fibrosis, and abundant hemosiderin are frequent findings. Histochemical staining is generally unnecessary for routine diagnosis of HH although Perls stain can help identifying hemosiderin deposits[1, 2, 5].

Treatment for HH is generally unnecessary due to its benign nature. Surgical excision may be considered for tissue diagnosis in uncertain cases, symptomatic lesions, or cosmetic reasons.

Recurrence and metastasis following curative procedures are rare[2].

In conclusion, HH presents with a diverse range of clinical and dermoscopic features, and the characteristic peripheral halo may be absent in many cases, reducing its diagnostic reliability. Clinicians should carefully consider differential diagnoses and rule out possible malignancies. Histopathological examination can provide a definitive diagnosis.

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References

1. Santa Cruz DJ, Aronberg J. Targetoid hemosiderotic hemangioma. J Am Acad Dermatol 1988;19:550-8.
2. Joyce JC, Keith PJ, Szabo S, Holland KE. Superficial hemosiderotic lymphovascular malformation (hobnail hemangioma): a report of six cases. Pediatr Dermatol 2014;31:281-5.
3. Porriño-Bustamante ML, Aneiros-Fernández J, Retámero JA, Fernández-Pugnaire MA. Hobnail Hemangioma With an Unusual Clinical Presentation. J Cutan Med Surg 2017;21:164-6.
4. Trindade F, Kutzner H, Tellechea Ó, Requena L, Colmenero I. Hobnail hemangioma reclassified as superficial lymphatic malformation: a study of 52 cases. J Am Acad Dermatol 2012;66:112-5.
5. Yoon SY, Kwon HH, Jeon HC, Lee JH, Cho S. Congenital and multiple hobnail hemangiomas. Ann Dermatol 2011;23:539-43.
6. Zaballos P, Llambrich A, Del Pozo LJ, et al. Dermoscopy of Targetoid Hemosiderotic Hemangioma: A Morphological Study of 35 Cases. Dermatology 2015;231:339-44.
7. Behera B, Chandrashekar L, Thappa DM, Gochhait D, Ayyanar P. Atypical clinical and dermoscopic features of hobnail hemangioma. Indian J Dermatol Venereol Leprol 2021;87:93-7.
8. Papageorgiou V, Apalla Z, Sotiriou E, et al. The limitations of dermoscopy: false-positive and false-negative tumours. J Eur Acad Dermatol Venereol 2018;32:879-88.
9. Enei ML, Paschoal FM, Valdes R. Arborizing vessels in a targetoid hemosiderotic hemangioma: mistaken dermoscopic diagnosis of basal cell carcinoma. Dermatol Pract Concept 2017;7:43-7.
10. Piccolo V, Russo T, Mascolo M, Staibano S, Baroni A. Dermoscopic misdiagnosis of melanoma in a patient with targetoid hemosiderotic hemangioma. J Am Acad Dermatol 2014;71:e179-81