Targetoid Hemosiderotic Hemangioma: A Case Report of Multiple Asymptomatic Lesions

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Practice Points

- Targetoid hemosiderotic hemangiomas (THHs) are benign lesions that typically are associated with trauma and may be influenced by estrogen levels.
- Treatment often is not necessary unless these lesions are symptomatic. No metastasis or systemic sequelae are known to occur with THHs.
- Targetoid hemosiderotic hemangiomas are now thought to be of lymphatic origin and typically stain with D2-40, vascular endothelial growth factor receptor 3, and the absence of CD34; however, CD34 may be positive if microshunts are present between THHs and blood vessels.

Targetoid hemosiderotic hemangioma (THH) is a benign vascular tumor characterized by a central violaceous papule with a clear periphery bordered by an ecchymotic ring. Originally coined by its characteristic halo appearance with hemosiderin deposits, not all THHs have this classic halo or hemosiderin composition. We report a unique case of THH in which the patient presented with multiple lesions with no prior trauma. Multiple THH lesions have been linked to minor trauma; however, the presence of 4 concurrent lesions with the absence of trauma makes this THH presentation atypical and unique.

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Case Report

A 47-year-old woman presented with multiple asymptomatic, nontender, erythematous to violaceous papules of 1 year's duration. The patient denied any trauma to the area. Her medical history was notable for asthma, vocal cord dysfunction, seasonal allergies, and asbestos exposure. Her family history was unremarkable.

Physical examination revealed 4 erythematous, slightly firm, nontender, nonblanching papules with surrounding rings of ecchymosis and hemosiderin deposition on the right abdomen (5×5 mm), right chest (6×4 mm), and bilateral upper legs (left medial thigh, 3×3 mm; right medial thigh, 5×7 mm) (Figure 1). No lymphadenopathy was noted.

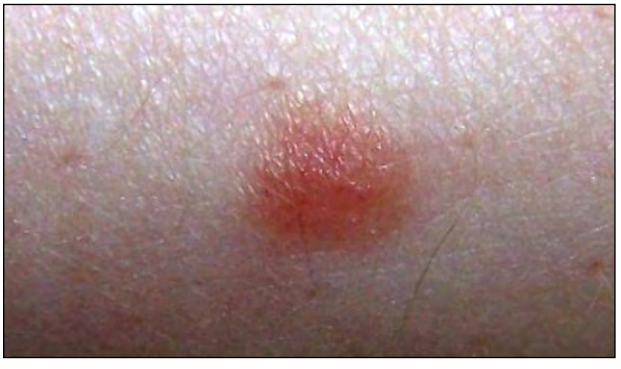

Figure 1. An erythematous papule with a surrounding ring of hemosiderin deposition on the abdomen (5×5 mm).

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A biopsy was performed for histologic examination and revealed dilated vascular structures in the upper dermis lined by prominent hobnail endothelial cells and collagen dissection (Figure 2). Narrow neoplastic vessels were present in the deeper part of the lesion with scant hemosiderin and erythrocyte extravasation. Despite the multiplicity of lesions, a diagnosis of targetoid hemosiderotic hemangioma (THH) was favored. The lesions were excised and similar histologic findings were confirmed. To date, the patient has reported no reoccurrences.

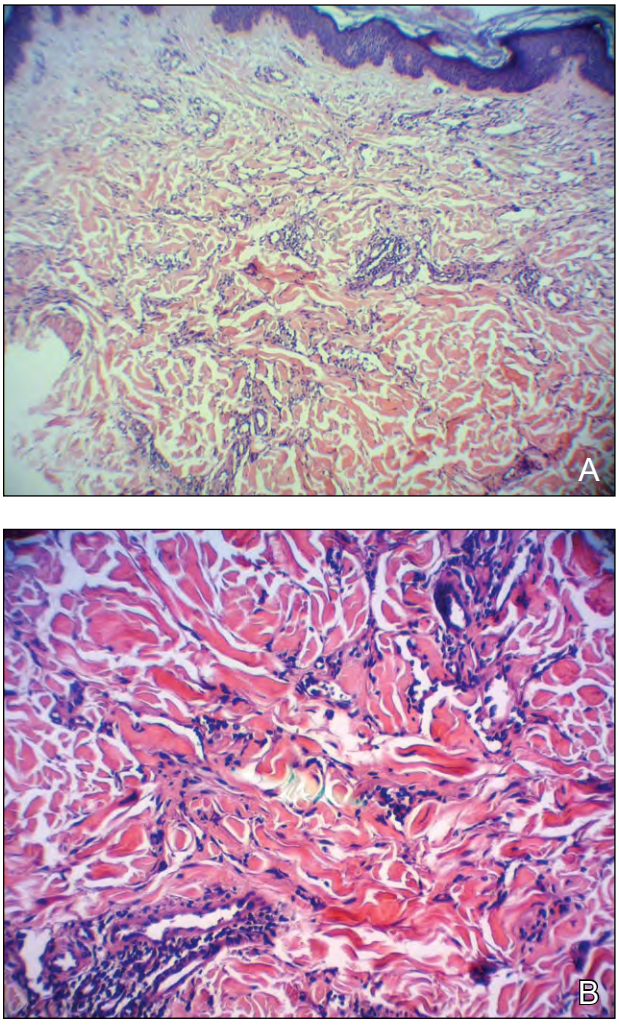

Figure 2. Histopathology of a biopsy demonstrated dilated vascular channels lined by epithelioid endothelial cells in the superficial portion of the lesion (A)(H&E, original magnification ×2.5). These cells formed intraluminal papillary projections. The deeper dermis exhibited much narrower, anastomosing, slitlike channels between collagen bundles. The vascular spaces in the early targetoid hemosiderotic hemangioma stage showed protrusion of plump endothelial cells, giving the lesion a characteristic hobnail appearance (B)(H&E, original magnification ×10).

Comment

Targetoid hemosiderotic hemangioma is a benign vascular tumor characterized by a central violaceous papule with a clear periphery bordered by an ecchymotic ring. Coined by its characteristic halo appearance,[1] not all THHs have this classic presentation.[2] Subsequently, *hobnail hemangioma* is a term used interchangeably to describe these lesions from the hobnail appearance of the endothelial cells. Targetoid hemosiderotic hemangioma has a biphasic growth pattern. In the initial stage, there is a solitary, red-blue, 2- to 3-mm papule with a clear periphery bordered by an ecchymotic ring. Classically, the ring will expand peripherally and eventually fade and disappear over time with retention of the central papule. These lesions also have been known to fluctuate with hormonal changes as seen in menstruation and pregnancy.[3] Estrogen is a vasoactive hormone that promotes vascular permeability and fragility, venous distensibility, vasomotor instability, and increased blood flow. Progesterone also has proven to have vasodilatory and vasoconstrictive effects on blood vessels.[4] The physiologic effects from hormonal changes observed in our patient, a 47-year-old perimenopausal woman, could explain the cause of these 4 lesions. Dermoscopy typically reveals well-demarcated red or reddish blue lagoons with few black macules representing hemorrhagic crusts. Fine delicate pigmentation may be found at the halo corresponding to hemosiderin deposition.[2]

Histologically, dilated vascular channels lined by epithelioid endothelial cells characterize the superficial portion of the lesion. These cells often will form intraluminal papillary projections. The deeper dermis exhibits much narrower, anastomosing, slitlike channels between collagen bundles. The vascular spaces in the early THH stage show protrusion of plump endothelial cells, giving the characteristic hobnail appearance.[5]

Although THH previously was thought to be a true vascular tumor, research has exposed a lymphatic origin from expression of D2-40 and vascular endothelial growth factor receptor 3. Researchers identified D2-40, a monoclonal antibody, as a marker expressed in lymphatic endothelial cells that is not present within blood vessel endothelial cells. Vascular endothelial growth factor receptor 3 is an immunohistochemical marker for lymphatic endothelial cells that is expressed in THHs and only weakly stains endothelial cells in cutaneous hemangiomas.[5] Furthermore, CD34, a marker for blood vessel endothelial cells, typically is absent within THHs.[6] However, when blood endothelial cells are histologically seen lying adjacent to the THH, CD34 has been expressed within the THHs. This marker indicates the presence of microshunts

between blood and lymphatic vessels in these lesions. These microshunts explain many of the histologic findings for THHs.[6]

The clinical differential diagnosis includes Kaposi sarcoma (patch stage), progressive lymphangioma, and angiokeratoma. Targetoid hemosiderotic hemangioma must be differentiated from other hobnail cytomorphologies, such as retiform hemangioendothelioma, acquired progressive lymphangioma, and papillary intralymphatic angioendothelioma.[6] Differentiation from these lesions requires identifying the presence of plump epithelioid endothelial cells with intraluminal papillary formations, hemosiderin depositions, scarcity of plasma cells, and cytoplasmic hyaline globules.[7]

Conclusion

Targetoid hemosiderotic hemangiomas are benign without any propensity for systemic metastasis. Treatment often is unnecessary, and a THH may fully regress without any treatment.[8] Our case of 4 lesions with the absence of trauma makes this THH presentation atypical and unique.

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