

Arborizing vessels in a targetoid hemosiderotic hemangioma: mistaken dermoscopic diagnosis of basal cell carcinoma

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ABSTRACT Targetoid hemosiderotic hemangioma (THH) or hobnail hemangioma (HH) is a benign vascular lesion that presents with the classical clinical presentation of a ring-shaped tumor having a targetoid appearance, with a central purple-brown papule surrounded by a thin pale area and an ecchymotic ring on the outside. Dermoscopic features and patterns of HH have been documented and have proven to be sufficient to establish a clinical diagnosis in many cases.

We present a facial lesion in which both the clinical presentation and dermoscopy were atypical. The presence of arborizing vessels in the dermoscopic pattern, never before described for this lesion, led us to the diagnosis of basocellular carcinoma (BCC). We also report the changes in this pattern experienced over 12 months of progression and their correlation with the histopathologic findings.

Introduction

Targetoid hemosiderotic hemangioma (THH) or hobnail hemangioma (HH) is a benign vascular lesion, which mainly affects young people with a prevalence in men.

The classical clinical presentation is a ring-shaped vascular lesion having a targetoid appearance, with a central purple-brown papule surrounded by a thin pale area and an ecchymotic ring on the outside [1,2]. However, HH tends to undergo cyclical changes and, therefore, the clinical presentation can vary. Thus, depending on the staging, the differential

diagnosis considered should include lesions such as hemangioma, angiokeratoma, Kaposi's sarcoma, dermatofibroma, insect-bite reactions, melanocytic nevus and melanoma.

Dermoscopy is a non-invasive diagnostic technique useful for diagnosing both melanocytic and non-melanocytic pigmented lesions, as well as vascular skin lesions. Dermoscopic features and patterns of HH have been increasingly documented and have proven to be sufficient to make a clinical diagnosis in many cases. Here, we present a facial lesion in which both the clinical presentation and dermoscopy were atypical. The presence of arborizing vessels in the dermo-

scopic pattern, never before described for this lesion, led us to the diagnosis of basal cell carcinoma (BCC). We also report the changes in this pattern experienced over 12 months of progression and their correlation with the histopathological findings.

Clinical Case

A 68-year-old man presented for consultation for a lesion on the nasal dorsum, which was asymptomatic with a few months of progression. There was no history of trauma or preexisting lesions in the area.

On clinical examination a pigmented papule was observed (5 x 5 mm) with a smooth surface and regular edges (Figure 1). Dermoscopic analysis with a FotoFinder video dermoscope (FotoFinder Systems GmbH, Bad Birnbach, Germany) detected the presence of arborizing vessels throughout the lesion, a bluish-gray ovoid structure, multiple cluster brown points and diffuse brown pigment (Figure 1).

These findings led us to the diagnosis of BCC and indicated resection. However, the patient returned after 12 months without presenting notable clinical changes. A new dermoscopy showed the presence of a dense bluish-brown pigment following a reticular distribution and the disappearance of most arborizing vessels; the extreme distal vessels remained in some areas and telangiectasia in others (Figure 2).

An excisional biopsy of the lesion was performed, and the tissue was sent for histopathologic analysis, which reported the presence of irregularly dilated blood vessels mainly in the superficial dermis having a prominent endothelium composed of relatively small cells, with little cytoplasm and hyperchromatic nuclei that projected towards the lumen (hobnail aspect). Also the presence of sparse hemosiderin and hemosiderotic histiocytes in the dermis was reported (Figure 3). The diagnosis was HTT. The patient has been managed for 10 years without further recurrence.

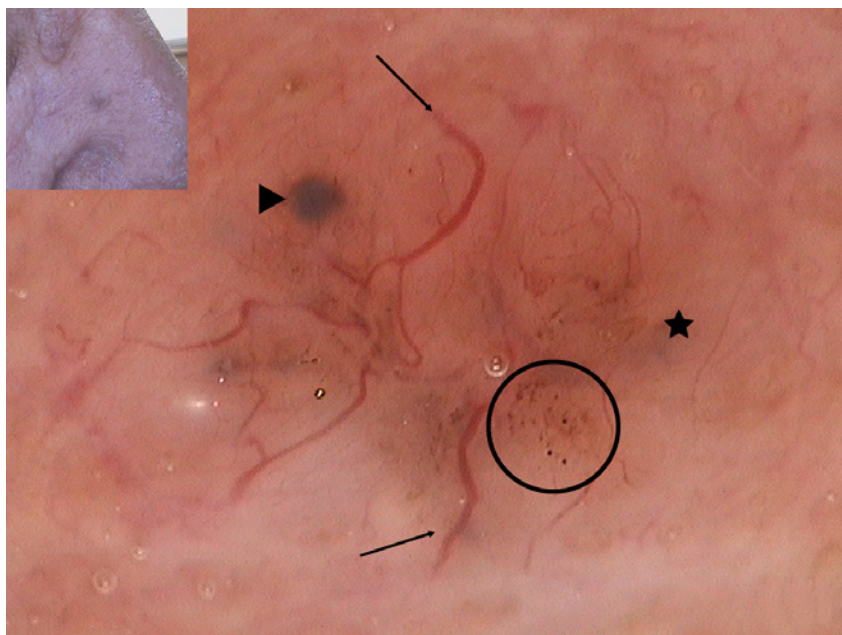


Figure 1. Clinical appearance. Pigmented papule, 5 mm diameter, nasal dorsum, brown to black, well delimited. The surrounding skin is of normal appearance. (b) Dermoscopic analysis: arborizing vessels (arrows), multiple brown points (circle) and a large blue-gray ovoid nest structure (triangle). There is little brown pigment dispersed through the lesion (star). [Copyright: ©2017 Enei et al.]

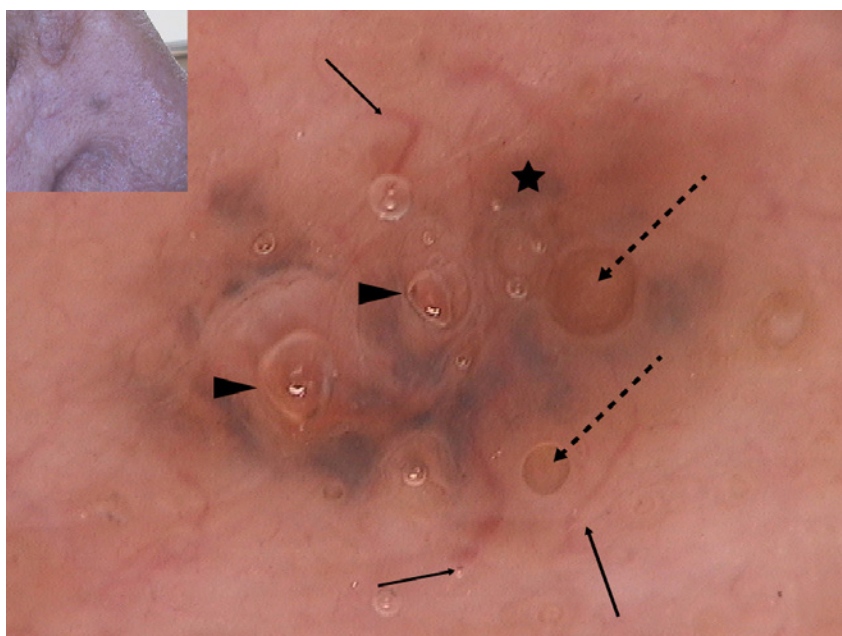


Figure 2. (a) Clinical appearance after one year of evolution. The papule shows no significant changes, including size and color (b) Dermoscopic analysis: the centre of the lesion displays a dense blue-brown pigment with reticular distribution (stars). At the periphery are the extreme distal vessels from Figure 1 (black arrows) and artifacts (black triangles). Two round, yellowish brown structures are in the middle of the pigmented area (segmented arrows). [Copyright: ©2017 Enei et al.]

Discussion

The histopathogenesis of HH is not yet clear. It is accepted that a local precipitating factor (trauma or hormonal influence) leads to the development of

microshunts between the lymphatic and blood vessels in the dermis. The capillary pressure of the latter would cause erythrocytes to pass into the lymphatic spaces forming aneurysmal structures. In older lesions, the emerging lymph

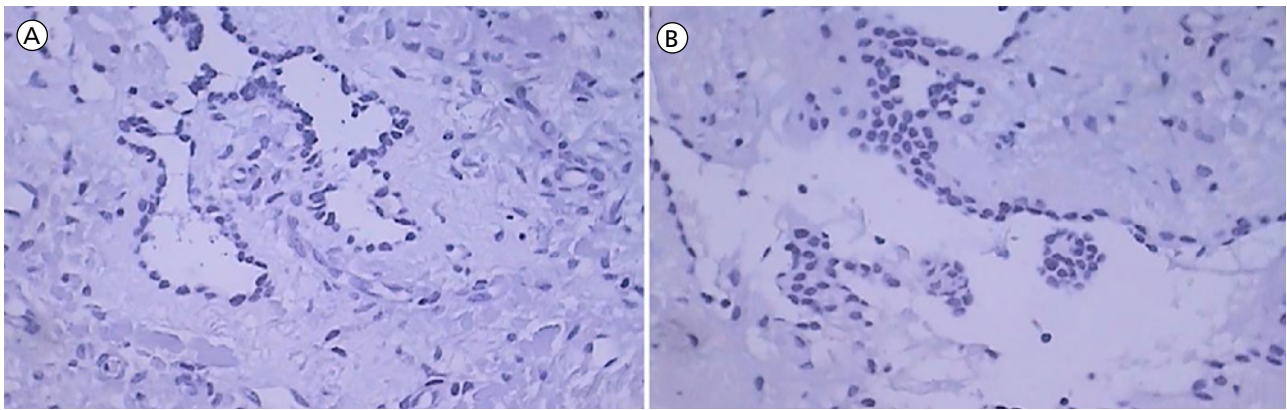


Figure 3. Histopathologic analysis. Irregularly dilated blood vessels mainly in the superficial dermis, having a prominent endothelium composed of relatively small cells (a), with little cytoplasm and hyperchromatic nuclei, projected towards the lumen (hobnail aspect)(b). [Copyright: ©2017 Enei et al.]

vessels become clogged, causing the disappearance of these ectatic vessels in the papillary dermis. Additionally, there is increased deposition of hemosiderin and subsequent fibrosis [3]. Thus, the histology shows a vascular lesion confined to the dermis, with a biphasic pattern. The upper portion has dilated and irregular vascular spaces with thin walls delineated by endothelial cells display a “hobnail” aspect, i.e., round, with little cytoplasm and a large nucleus that protrudes into the vascular lumen. In the lower portion, thinner vascular spaces appear to cut their way through the collagen bundles. Local deposits of hemosiderin or siderophages are common, secondary to a local hemorrhagic processes.

Although studies based upon immunohistochemistry lead to recognition of the lymphatic origin of HH [4], recent observation of these markers suggests also a biphasic histological behavior for these lesions. Takayama, et al [5], found not only positivity for CD31 and D2-40 in the upper portion of the hobnail, confirming the lymphatic nature of the lesion, but also positivity for CD31, CD34-factor VIII, and α -SMA, suggesting an endothelial origin of the lower portion. As far as diagnosis is concerned, this is often difficult, especially if the lesion lacks the typical targetoid appearance.

Dermoscopy is a noninvasive imaging technique that allowed us to significantly increase the diagnostic precision

for skin lesions. The first reference to epiluminescence microscopy for HH dates back to 1998 and describes well-demarcated red lagoons in the papular region of the lesion [6]. Since then, no more than 12 reports based on this technique have been published, covering a total of 48 lesions studied (Table 1; [5-16]).

We highlight the series of 35 cases recently published by Zaballo et al [7] in which the authors were able to establish that the unequivocal dermoscopic pattern for HHT is only present in 52% of lesions and is represented by the presence of lagoons in the center, a yellowish circular intermediate area, and a purple or ecchymotic ring on the periphery.

In this series, the most common pattern (71.2%) consisted of central lagoons and a homogeneous area on the periphery, which can also be observed in other vascular tumors and 22.8% of the lesions were fully occupied by a reddish purple or reddish brown homogeneous area.

This variety of dermoscopic patterns correlates with the different stages of tumor progression. Thus, the proliferation of dilated vessels in the superficial dermis correlates with the lagoon structures; the hemorrhagic phenomena are reflected in the ecchymotic ring, and fibrosis of older lesions gives rise to whitish homogeneous structures. In our case, neither of the two dermoscopic evaluations showed the structures or patterns described to date for HH. Thus,

we highlight that, as far as we know, this is the first time these arborizing vessels have been mentioned for this lesion. This finding, in conjunction with the blue, round structure, was the pattern that led us to posit the diagnosis of BCC.

However, several lesions have been reported to predominantly feature arborizing vascular structures, making it impossible to discard the diagnosis of BCC. Some examples are hidradenoma, trichoblastoma, trichoepithelioma, cylindroma, hydrocystoma, and poroma.

Other vascular structures, such as points or polymorphous irregular vessels, have been described in HH in the central portion of the lesion, which have been linked to deeper vessels in the dermis [7].

On the other hand, we believe that the round, blue structure and brown points, in addition to the diffuse brown pigmentation of the first image (Figure 1), corresponded as a set to an ecchymotic point and initial deposition of hemosiderin and siderophages in the middle dermis, respectively; this phenomenon increased until becoming a dense network in the center of the lesion after 12 months of progression, covering the globular structure and brown points.

It was interesting to compare the two dermoscopic studies and observe the collapse of the central portion of the vessels with the persistence of extreme fine vessels and telangiectasias. The emergence of two round, yellowish brown

TABLE 1. Published Dermoscopic Descriptions of Hobnail Hemangioma

Year published	Reference number	Number of cases	DC pattern	Vessels	HP	Dermoscopy monitoring	Clinical and DC diagnosis
2016	8	1	B	No	No	Yes	Yes
2015	7	35	A (71.4%) B (52%) C (22.8%)	Dots and polymorphous (40%)	Yes	Not described	Yes (77%)
2015	9	1	D	No	Yes	No	No
2015	10	1	A	No	No	Yes	Yes
2015	5	1	B	No	Yes	Yes (4 weeks)	Yes
2014	11	1	E	No	Yes	No	No
2010	12	1	A	No	Yes	No	No
2009	13	1	D	No	Yes	No	No
2009	14	1	A	No	No	Yes (3 months)	Yes
2007	16	1	A-D	No	Yes	No	No
2005	16	3	B	No	Yes	Yes	Yes
1998	6	1	A	No	Yes	Yes (34 days)	Yes

A: Central red and dark peripheral circular lacunae and reddish-violaceous homogeneous area / violaceous nuclei surrounded by a pale halo / well-defined network of circular blebs.

B: Red lacunae located at the center, an intermediate yellow circular homogeneous area, and a violaceous or ecchymotic homogeneous ring on the periphery. C: Homogeneous area.

D: Red, roundish lagoon-like areas in the focus of the injury, with a fine pigment network at the periphery. E: Slight peripheral pigment network surrounding an extensive reddish, structureless area with chrysalis-like structures. HP: Histopathology. DC: Dermoscopy.

structures in the middle of the pigmented area is also reported (Figure 2), which could correspond to ectatic vessels in the superficial dermis described in the histopathology report, similar to the structures observed in the lymphangiomas. In neither of the two stages did we observe homogenous areas, lagoons, thin pigmented networks, or whitish structures, such as the ones described in published cases of HH.

In conclusion, arborizing vessels and blue clods are not unique to BCC and have been described for other tumors. However, this is the first time that they are described in a lesion of HH. We highlight this case because of its unusual dermoscopic presentation. Therefore, we add BCC to the differential diagnosis of HH and highlight the fact that when the clinical presentation is uncharacteristic, histopathology continues to be the gold standard for diagnosing this lesion.

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