

## A Recurrent Case of Targetoid Hemosiderotic Hemangioma: A Case Report and a Comprehensive Review of the Literature

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**ABSTRACT** Targetoid hemosiderotic hemangioma is an acquired vascular malformation of unknown origin. We report the case of a 31-year-old man with a recurrent and spontaneous regressive targetoid hemosiderotic hemangioma. Diagnosis relied on clinical and histological findings. Physical examination revealed presence of an approximately 2 cm targetoid lesion located on the left arm, and associated with pain after pressure. No trigger agent (trauma, insect sting) was reported. Dermoscopy showed a group of red lacunae centrally, encircled by an intermediate yellow circular homogenous area and a red violaceous homogenous ring in the periphery. The histopathological examination and the immunohistochemical staining of the lesion were characteristic for a hemangioma-like proliferation of vessels in the upper part of the dermis, similar to a targetoid hemosiderotic angioma. We also review epidemiological, clinical, and histopathological findings in 6 similar cases presented in the literature. Spontaneous regression and recurrence have rarely been described in this type of skin lesion.

**KEY WORDS:** targetoid hemosiderotic hemangioma, recurrent, regressive

### INTRODUCTION

Targetoid hemosiderotic hemangioma (hobnail hemangioma) is a rare benign vascular tumor described for the first time in 1988. The female-to-male ratio reported in the literature is 1:1, with an age of onset ranging from 5 to 67 years (1).

The etiology of this disease is incompletely elucidated. A commonly accepted theory is that it may occur on a pre-existing hemangioma, after a trauma. Trauma could induce the formation of thrombi and microshunts, with pressure from the capillaries causing the filling of the lymph spaces with erythrocytes, contributing to the formation of aneurysmal micro-

structures. The obstruction of some lymphatic vessels could induce inflammation, fibrosis, and interstitial hemosiderin deposits (2). The targetoid appearance could be explained by hemorrhage from the vascular proliferation, causing a purpuric ring with hemosiderin deposits.

The diagnosis relies on clinical and histological findings. Clinically, hemosiderotic hemangioma typically appears as a solitary small targetoid lesion with a central red/ purple and/or brown papule, encircled by a pale area and peripheral ecchymotic ring. It is usually located on the limbs and trunk, but unusual

presentation on the scalp has also been described (3). Clinical differential diagnosis includes Kaposi's sarcoma, hemangioma, dermatofibroma, melanocytic nevus, insect bite reaction, and solitary angiokeratoma (4).

Dermoscopy most frequently shows centrally located red lacunae and a homogenous red-violaceous area in the periphery. Some lesions may also present a delicate pigmented network in the periphery (5).

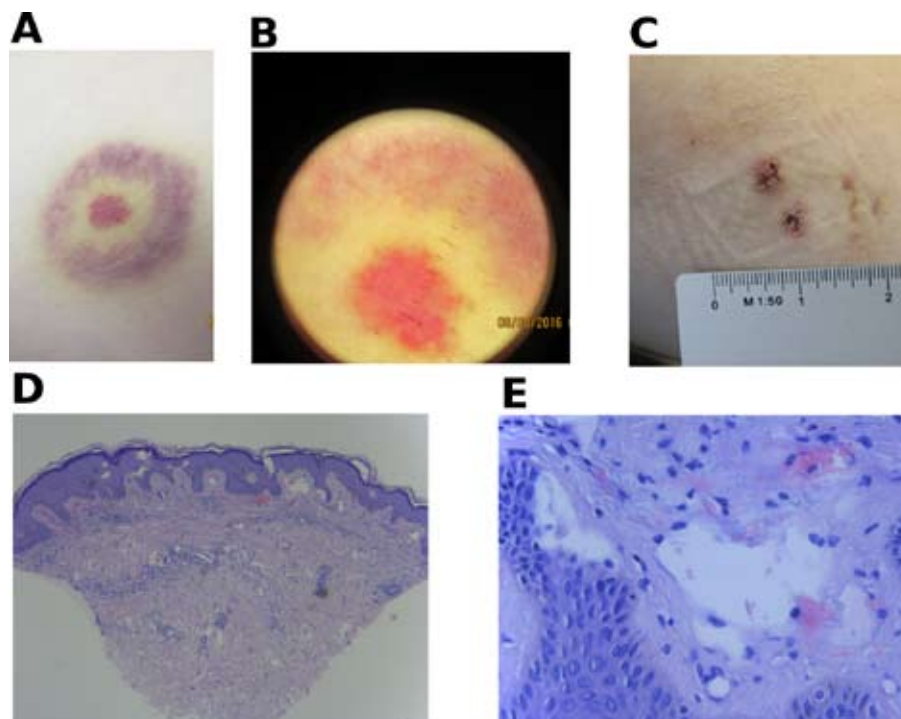
Histologically, hemosiderotic hemangioma presents a non-circumscribed dermal vascular proliferation with thin-walled dilated vessels in the papillary dermis continued with narrower, slit-like endothelial lined spaces in the deep reticular dermis, dissecting between dermal collagen bundles. Extravasated erythrocytes and hemosiderin deposits can be observed in the dermis. An inflammatory lymphocytic interstitial or perivascular infiltrate is often seen. The lymphatic origin of targetoid hemosiderotic angiomas has been confirmed by recent studies, which demonstrated the positive immunohistochemical staining of these lesions for CD31, vascular endothelial growth factor receptor-3 (VEGFR-3), the lymphatic specific antipodoplanin monoclonal antibodies (D2-40), and also a lack of CD34 staining. The absent or minimal proliferative nature of this vascular lesion has been suggested by a consistent very low Ki-67 proliferation index. The histopathologic differential

diagnosis includes patch stage and lymphangioma-like variants of Kaposi's sarcoma, well-differentiated angiosarcoma, retiform hemangioendothelioma, Dabska tumor, and benign lymphangiomas. In contrast to Kaposi sarcoma, targetoid hemosiderotic angioma does not stain positively for human herpesvirus 8 (6).

The evolution of this benign tumor is different. It is permanent in most of the cases, and it is generally accepted that it does not resolve itself. An excision may be indicated, if the lesion is cosmetically disturbing (7). To our best knowledge, there are only a few case reports of targetoid hemosiderotic hemangioma with recurrent evolution in the literature (4,7,8).

### CASE REPORT

A 31-year-old male patient from an urban area presented in our department in August 2016, with a 1-week history of spontaneous occurrence of a targetoid lesion, located on the left arm (Figure 1, A). He also reported pain on pressure. The patient reported that the lesion had occurred one year before, approximately 2-3 times, in the same place. The lesion had an approximate duration of 10 days and then disappeared completely between cycles. The skin in the region looked normal, without evidence of a papule, postinflammatory hyperpigmentation, or scarring. Personal medical history was not significant.



**Figure 1.** Clinical, dermoscopy, and histopathological features of our patient at diagnosis. (A) Clinical aspect at diagnosis. (B) Dermoscopy at diagnosis. (C) Clinical aspect after regression. (D) Histopathological aspect, low magnification. (E) Histopathological aspect, high magnification. Hobnail endothelial spaces in the center of the lesion.

Physical examination revealed the presence of an approximately 2 cm targetoid lesion with a central red papule, encircled by a pale area and a peripheral ecchymotic ring, located on the left arm. Dermoscopy showed a group of red lacunae centrally, encircled by an intermediate yellow circular homogenous area and a red violaceous homogenous ring in the periphery (Figure 1, B).

Histopathologic examination was performed on two samples. The histopathological examination of the sample from the middle of the lesion showed a slightly irritated epidermis. In the dermis, we observed slit-like proliferations of vessels, with prominent endothelium in some places, erythrocytes, a sparse inflammatory infiltrate, and few plasma cells (Figure 1, D, E). The histopathological examination of the sample from the margin showed a very sparse finding, with few erythrocytes and few prominent vessels. The immunohistochemical staining of the sample from the middle showed positive staining of the vessels from the dermis for CD31, positive staining of some vessels for D2-40 (podoplanin), and negative staining for HHV8. Actin (alpha smooth muscle) staining showed muscular differentiated cells around the vessels. MIB1 staining showed no obviously proliferating endothelium. Therefore, the lesion was evaluated as a hemangioma-like proliferation of vessels in the upper part of the dermis. The histology was similar to a targetoid hemosiderotic angioma.

No local or systemic treatment was administered. The hemangioma completely disappeared without leaving any scar a few days after the punch biopsy from the middle and margin of the lesion was performed (Figure 1, C).

The patient was contacted after 22 months. He reported no new occurrence of the lesion.

## REVIEW OF LITERATURE

We have also performed a review of the literature. The PubMed database was searched for the following terms: "targetoid", "hemosiderotic", "hemangioma", "recurrent", "regressive".

We reviewed a total number of 6 cases with recurrent targetoid hemosiderotic hemangioma. The clinical and histopathological features of the patients are shown in Table 1.

## DISCUSSION

Targetoid hemosiderotic hemangioma may represent a transient inflammatory phase in the natural evolution of a capillary hemangioma (4). These lesions generally persist, and no treatment is recommended.

Excision is recommended in case of esthetic issues. Spontaneous total regression and cyclic changes, such as swelling and intermittent changes in lesion size and rim without complete regression, have been reported in association with menstruation (4) or in children (6).

Regression represents a common feature in infantile capillary hemangiomas. This event is supposed to be caused by apoptosis, as a higher number of apoptotic cells was found in these hemangiomas, compared to lobular capillary hemangioma. Regression was also reported in tufted angiomas (9).

We report the case of a 31-year-old man with a recurrent targetoid hemosiderotic angioma, appearing and disappearing in the same place without leaving any scars. The patient reported 2-3 similar episodes in the past year. No trigger agent (trauma, insect sting) was reported.

To our best knowledge, there are only 6 cases of recurrent targetoid hemosiderotic hemangioma reported in the literature (see Table 1). The age of the patients ranged between 9 and 40 years. Trauma (bee sting) was incriminated as a potential trigger in only one patient (case 5) (Table 1). Occurrence at menstruation was mentioned in the same patient (case 5) (Table 1). The lesions were situated on the trunk in 2 patients (case 2 and 5), upper limbs in 3 patients (case 1, 3, and 6) and lower limbs in 1 patient (case 4). Clinically, all lesions presented a central papule with an ecchymotic ring in the periphery in 3 patients (case 1, 4, 5), an erythematous ring in 1 patient (case 2), and a brown-gray macule in 2 patients (case 3 and 6). As for associated symptoms, itching and burning were present in one patient (case 2). The other patients reported no associated subjective symptoms. In histopathological examination, the patients demonstrated histopathological characteristics already described in previous publications, as mentioned in Table 1. We could not identify any specific epidemiological, clinical, or histopathological findings that could explain the recurrence of the lesions in the 6 reported cases.

There are a few speculative theories on recurrence of such lesions in the literature. The cyclic changes related to menstruation could be the result of fluctuation of levels of circulating estrogen, which could cause instability of vessels and leakage (4). Another possible explanation could be trauma. None of these factors were identified in our patient. Additionally, a statistically significant larger size has been reported in targetoid hemosiderotic hemangiomas with episodic changes compared with persistent lesions (4).

Therefore, the reason for the recurrence and cyclic changes of this fascinating lesion remains a mystery.

**Table 1.** Previous cases of recurrent targetoid hemosiderotic hemangioma.

Case	Sex	Age	Trigger	Localization	Duration until spontaneous remission	Clinical morphology	Associated subjective symptoms	Histopathological findings
1 (7)	M	9	No trauma	Left shoulder	3 months	1.7 cm targetoid violaceous papule with ecchymotic ring	No	Punch biopsy Superficial dermis: dilated and proliferated capillaries, with hobnail endothelial cells protruding into the lumen, parallel to the skin surface. Congested highly dilated lumens filled with eosinophilic, homogenous materials. Mid-dermis: slit-like vessels dissecting between collagen bundles. Wide amounts of extravasated erythrocytes and hemosiderin deposition in the dermis.
2 (8)	M	26	No trauma mentioned	Right-sided flank	Recurrence in cycles every 4 to 5 months in the last past 4-5 years	6 mm violaceous papule with a surrounding 1.5 cm erythematous annular halo	Itching and burning	Removal of the lesion by elliptical excision. Detailed histopathology not available.
3 (4)	M	21	No trauma	Proximal right forearm below the elbow	Recurrence in cycles every few months in the last past 4 years	Tan-gray, 1 cm macule with a central 3 mm papule	Not mentioned	Removal of the lesion by elliptical excision. Epidermal hyperplasia. Dermis: wedge-shaped vascular tumor with vascular ectasias, slit-like spaces, lymphangiectasias and peripheral hemosiderin deposition
4 (4)	M	16	Not mentioned	Knee lateral to the patella	2 cycles in the last 2 years	Enlarging, non-tender 9 mm, black papule with an ecchymotic halo	Not mentioned	Removal of the lesion by elliptical excision. Diagnostic features of a targetoid hemosiderotic hemangioma and a foreign body type giant cell reaction
5 (4)	F	40	Bee sting	Left side of the upper back	No changes in pregnancy. Cyclical changes returned with resumption of menses	Appearance changed from a white-tan 5 mm papule to a deep black 14 mm papule with an ecchymotic ring every other menses	No	Removal of the lesion by elliptical excision. Features of a targetoid hemosiderotic hemangioma. Presence of the "promontory sign" (lymphangiectasia containing island of normal vessels surrounded by numerous telangiectasias)
6 (10)	M	12	Not mentioned	Left forearm	History of involution with recurrence	2 mm red vascular papule with surrounding brown-gray macule	No	D2-40 positive

In our opinion, physicians should be aware of the presence of this rare clinical entity and include it in the differential diagnosis of vascular lesions, including pediatric cases.

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