A Recurrent Case of Targetoid Hemosiderotic Haemangioma

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Sir,

Targetoid hemosiderotic haemangioma (THH) was first described in 1988 by Santa Cruz & Aromberg (1) as a dermatosis of a small benign vascular tumour of the superficial and mid-dermis. It is characterized by the appearance of a brown-to-violaceous central papule, surrounded by a thin, pale area and a peripheral ecchymotic halo. It is generally accepted that THH does not resolve itself. There is no recurrence following excision, although episodic and cyclic morphological changes have been recorded (2). Since its first description, more than 158 cases have been documented, all in Caucasians (3). We report here a case of THH in China that recurred following previous spontaneous resolution.

CASE REPORT

A 9-year-old Chinese boy presented approximately one month after the sudden appearance of an asymptomatic, violaceous papule on his left shoulder, followed by a persisting ecchymotic ring. There were no subjective symptoms. He had described a similar onset of the lesion one year previously, which had resolved completely and spontaneously within 3 months with no treatment. On examination, a 1.7-cm targetoid violaceous papule with an annular violaceous ring was noted on his upper left shoulder (Fig. 1). Punch biopsy and histological evaluation revealed dilated and proliferated capillaries in the superficial dermis, with hobnail endothelial cells protruding into the lumen. The highly dilated lumens were congested with eosinophilic, homogenous materials. These blood vessels or lymphatic vessels run parallel to the surface of skin. Slit-like vessels were located in the mid-dermis and dissected between collagen bundles. Extravasated erythrocytes and haemosiderin deposition were widely present in the dermis (Fig. 2 a, b). Smaller vessels were present, dissecting through collagen in the mid-dermis, with prominent dermal haemosiderin deposition (Fig. 2 c). The patient did not report any history of trauma to the pre-existing lesions in the area. All family members were Caucasian, and there were no relatives of Chinese origin. There were no systemic symptoms and the patient appeared to be in good general health.

On the basis of the clinical and histological features, a diagnosis of THH was made.

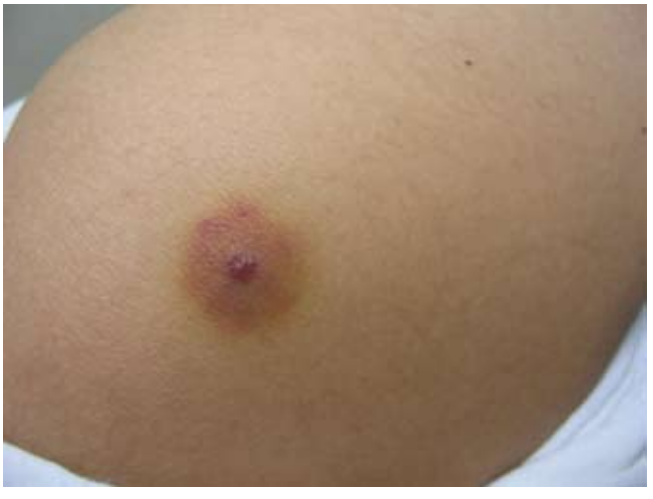

Fig. 1. A 17-mm diameter, targetoid violaceous papule with an annular violaceous ring on the upper area of the left shoulder.

DISCUSSION

THH, also known as hobnail haemangioma, is a benign, solitary, targetoid vascular neoplasm occurring predominantly on the proximal extremities and trunk, although facial locations have been recorded (1, 3). Published reports of THH reveal an equal gender incidence, with an age range of presentation of 5–67 years. THH is identified more frequently in younger persons (1).

Most patients exhibit a typical, targetoid haemangioma, whereby a small, solitary, purple and/or brown papule is surrounded by an ecchymotic ring that can expand or subsequently disappear with persistence of the central papule. The halo, however, is not a constant finding, thus the term hobnail haemangioma was coined to describe the non-targetoid variant of THH (4). Duration of the lesions ranges from 1 month to 20 years. During this time, THH may undergo episodic and cyclic changes in morphology (2). Complete and spontaneous clearing, as in our patient, has not been reported previously. Vascular proliferation in THH usually assumes biphasic growth patterns: the centre of the lesion is composed of dilated, irregular, thin-walled, ectatic, vascular spaces positioned in the superficial plexus. These vascular spaces are commonly lined with prominent plump endothelial cells protruding into the

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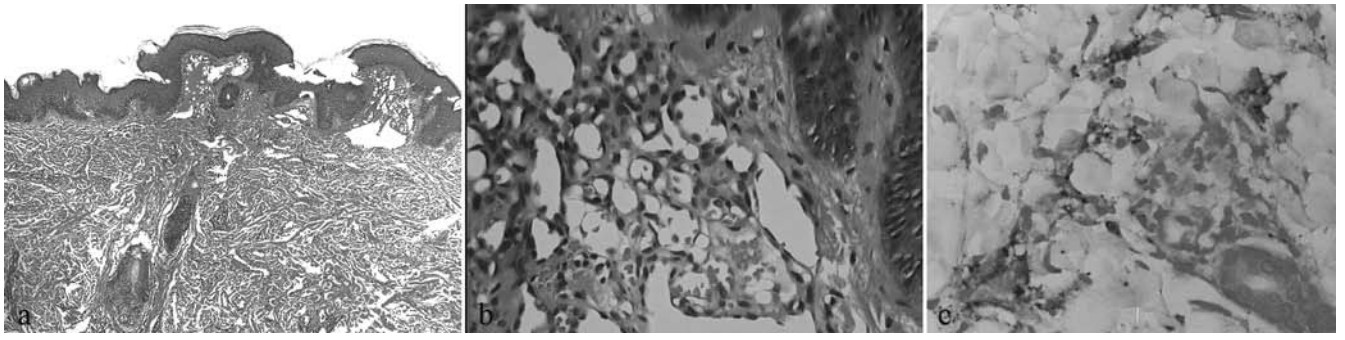

Fig. 2. (a) Histological evaluation revealed dilated and proliferated capillaries in the superficial dermis. Some highly dilated lumens were congested with eosinophilic, homogenous materials. Blood vessels or lymphatic vessels run parallel to the skin surface. Slit-like vessels were located in the mid-dermis and dissected between collagen bundles (H&E ×100). (b) Vessels were lined with plump, hobnail-like endothelial cells protruding into the lumen. Extravasated erythrocytes were obvious (HE ×400). (c) Staining for iron highlighted abundant haemosiderin deposition between collagen bundles (Prussian blue ×400).

lumen of the vessels. Whereas vessels in the middle or deep dermis shows irregular, angulated, thin-walled, slit-shaped vascular channels that dissect between collagen bundles of the dermis. Haemosiderin deposits, extravasated erythrocytes and a mild mononuclear inflammatory infiltrate are frequent findings (5, 6). Pathological changes in our case were basically coincident with those findings in THH. However, our patient had a more prominent telangiectasia and lymphangiectasis coexisted in the superficial dermis, which favours the recent notion that THH is a lymphangiectasia, not merely a dilation of blood vessels. Immunohistochemical study demonstrated that the neoplastic endothelial cells in THH are strongly positive for CD31, and rarely stain for CD34. Fifty percent of studied cases have been shown to express VEGFR-3, which suggests a lymphatic line of differentiation in hobnail haemangioma (4).

THH should be differentiated from patch-stage Kaposi's sarcoma (KS). In addition to having different clinical characteristics from patch-stage KS, THH usually displays widely dilated vascular spaces in the superficial dermis, a feature not found in patch-stage KS (7). Well-differentiated angiosarcoma, THH and retiform hemangloendothelioma (RHE) may all present proliferations of endothelial cells that have hobnail morphology. In this case, the clinical presentation is of importance. Cutaneous angiosarcoma occurs mainly in the head and neck region of elderly patients, and is characterized pathologically by anastomosing vascular structures lined by atypical and proliferative active endothelial cells. RHE presents as a plaque-like vascular mass. Its morphological hallmark is long, arborizing, thin-walled vascular spaces that infiltrate in a retiform pattern reminiscent of the normal rete testis (8).

Trauma is the only known predisposing factor, for example due to irritation from a belt or insect bites. Episodic changes in the form of an enlarging and diminishing haemorrhagic/hemosiderotic halo also implicate recurring trauma or vessel fragility (9). Hormones may also influence the clinical morphology, resulting in the cyclic changes of waxing and waning diameter and peripheral colour (3). The case described here had none of these potential triggering factors.

There is no effective preventative treatment for THH. Excision of the lesions may have a favourable prognosis with no malignant outcome or relapse.

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