

## Hemoporphin photodynamic therapy for a case of unilateral nevoid telangiectasia

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*To the Editor:* Unilateral nevoid telangiectasia (UNT) is an uncommon cutaneous vascular disorder characterized by superficial telangiectasia distributed in a unilateral, linear pattern. It can be either congenital or acquired. The pathogenesis of UNT is unclear and most scholars thought that hyperestrogenic states play an important role in UNT. Pulsed-dye laser (PDL) therapy is a mainstream treatment option. Herein, we present a case of PDL-resistant UNT successfully treated with hemoporphin-mediated photodynamic therapy (HMME-PDT).

An 8-year-old boy presented with a 4-year history of progressive asymptomatic erythema. The lesion was fingertip size at the beginning, only affected the extensor aspect in the left lower leg, gradually enlarged and spread to the entire left lower extremity [Figure 1A]. The lesion blanched completely by diascopy. He had experienced two times of 585 nm PDL therapy but without evident improvement. There was no medical history of hepatic disease or other diseases. A general examination was unremarkable. Examination revealed generalized telangiectasia along the unilateral body in a linear distribution, involving the left lower extremity, the left buttock, and the left back. Laboratory tests revealed no abnormalities in blood routine examination, liver and renal function, blood coagulation, and sexual hormones. Dermatoscopy showed branched vessels with a reddish background [Figure 1B]. A skin biopsy from the left leg revealed normal epidermis and dilated capillaries with little erythrocyte exosmosis on the superficial dermis [Figure 1C]. The deformed vessel walls stained positive for a vascular endothelial growth factor (VEGF) [Figure 1D], but negative for podoplanin (D2-40), glucose transporter-1 (Glut-1), and estrogen receptor/progesterone receptor (ER/PR).

Based on these findings, a diagnosis of UNT was made. After obtaining the patient's parental consent, HMME-PDT was taken to treat the lesions. Setting the larger spot

(diameter: 10 cm) above the left shank, the smaller one (diameter: 4.5 cm) above the left thigh, the patient was irradiated with 532 nm LED green light equipment for 20 min after intravenous infusion of HMME (5 mg/kg). The power density is 90 mW/cm<sup>2</sup>. The lesion resolved significantly after 3 months of follow-up. In the second session, the patient's parents requested enlarging the exposure area to save treatment costs. After signing the informed consent form, two sets of equipment were used for irradiation under close clinical monitoring [Figure 1E]. The obvious removal of lesions (>85%) was observed after the second session, with only slightly local skin hyperpigmentation [Figure 1F]. No abnormalities of blood routine examination, coagulation parameters, and liver or renal function were found during the follow-up. The dilated blood vessels were significantly reduced under dermatoscopy [Figure 1G]. No other adverse reactions and recurrence were observed within 14 months of follow-up.

The estrogenic role in the pathogenesis of UNT remains equivocal because several patients showed normal estrogen and progesterone levels and the lack of ER/PR in the skin. The predilection sites of UNT are C3–C4 and trigeminal nerve dermatomes. Only about a dozen cases have been reported involving lower limbs till now.<sup>[1,2]</sup>

The main clinical differential diagnosis of UNT includes telangiectasia macularis eruptiva perstans (TMEP), angioma serpiginosum (AS), and port wine stains (PWS). An increased number of mast cells in the superficial dermis of TMEP by Giemsa staining can distinguish it from other diseases. AS occurs more frequently in young women. Clinically, AS is typically distributed in a linear, gyrate, or serpiginous pattern. Unlike UNT, AS does not blanch by diascopy, and under dermatoscopy the existence of multiple oval red lagoons is characteristic. Histologically, the endothelial cells of AS are thick-walled without erythrocyte

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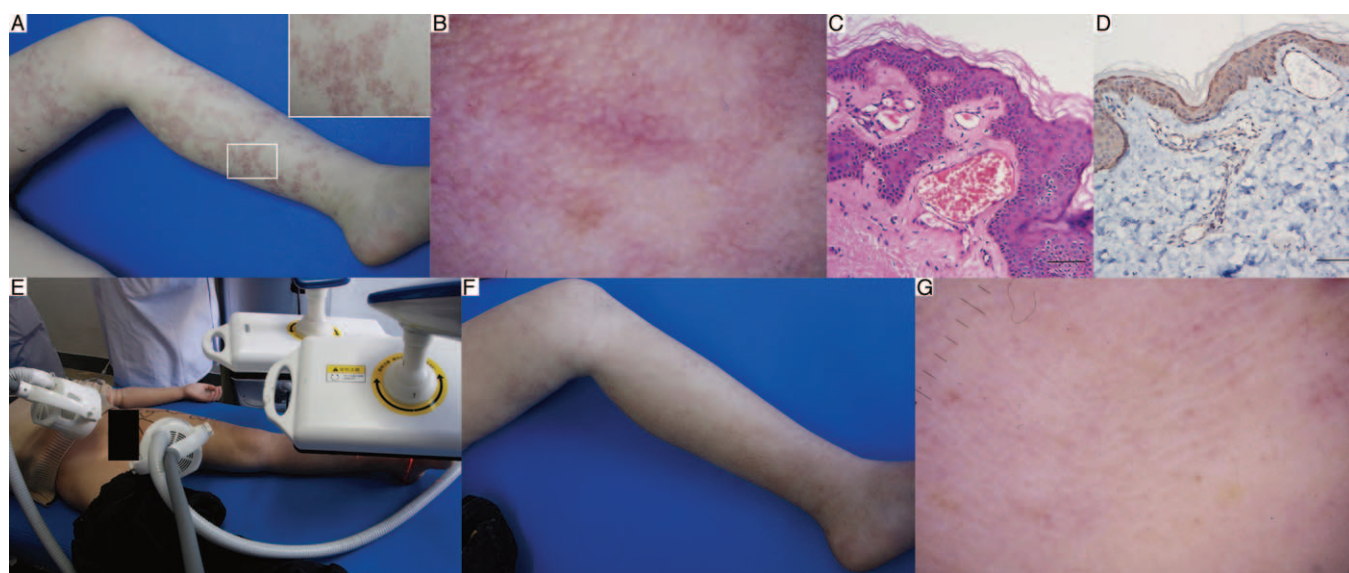
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**Figure 1:** Clinical, histopathological, and dermoscopic features of the patient. (A) A pre-treatment close-up of the multiple telangiectases distributed in the left lower extremity. (B) Dermoscopy showed branched vessels with a reddish background before treatment. (C) Histopathological findings of the lesion from the left leg showed normal epidermis and dilated capillaries with little erythrocyte exosmosis on the superficial dermis (H&E). Bar = 100  $\mu$ m. (D) The endothelial cells of deformed vessel walls stained positive for VEGF. Bar = 100  $\mu$ m. (E) The patient was irradiated with two sets of 532 nm LED green light equipment in the second treatment (Immunohistochemistry). (F) The obvious removal of lesions was observed after two sessions. (G) Dermoscopy revealed less branched blood vessels and clear background after two sessions. VEGF: Vascular endothelial growth factor.

extravasation. As for PWS, it usually appears after birth and appears as a more macular clinical manifestation with clear boundaries, gradually getting deeper in color and thickening in-depth with age, even evolving into nodular hyperplasia; The area of lesions enlarges proportionally with physical growth and commonly does not incline to the peripheral extension. Pathologically, the dilated vessels can reach the lower dermis and subcutaneous tissue as the disease progresses, while the telangiectasia of UNT is confined to the superficial and mid-dermis.

In patients with high estrogen levels, the telangiectasia may fade when estrogen returns to normal; However, in an overwhelming majority of UNT cases without hormonal abnormalities, symptomatic treatment can meet the needs of cosmetic improvement. PDL has been employed to treat UNT for many years. Yellow light (*eg.* 585 or 595 nm) produced by PDL can be absorbed by hemoglobin, causing a blanching response through complete photocoagulation. Some UNT patients can benefit from PDL. However, it also has several limitations. For example, the competitive absorption of epidermal melanin to light in this spectral range attenuates the efficiency of PDL treatment. Second, PDL treatment only affects the blood vessel with a diameter of 50 to 150  $\mu$ m. Third, PDL has a good response to dotted and globule patterns while it is not effective for reticulated blood vessels under dermoscopy. Besides, Sharma and Khandpur<sup>[2]</sup> reported that only a moderate response (26–50% lightening) has been achieved in UNT patients after an average of 2.33 sessions of PDL treatment. Moreover, Cliff and Harland<sup>[3]</sup> have previously treated five cases of UNT with PDL, all of which showed evidence of recurrence. We speculated that the patient's resistance to PDL may be due to the great vessel diameter (>150  $\mu$ m as shown in Figure 1C) and the branched vessels.

In 2016, HMME-PDT was approved for its application in treating PWS in China. It performed excellently in the treatment of PWS, especially in PDL-resistant PWS.<sup>[4]</sup> PDT uses the cytotoxic singlet oxygen produced by the interactions between light, photosensitizers, and oxygen to damage the deformed capillary network, cause cell death, endothelial damage, thrombosis, and vascular occlusion.<sup>[4,5]</sup> Compared with PDL, PDT can selectively target dilated vessels of any size and penetrate deeper. Besides, PDT can destroy dilated vasculature through intracapillary photochemical and photothermal reactions simultaneously, but PDL only works through photothermolysis.<sup>[4]</sup> Moreover, the normal epidermis and dermal tissues can escape the damage from PDT since the low concentration of photosensitizer in them, but high in the endothelial cells.<sup>[5]</sup> Therefore, HMME-PDT is a safe method for PWS with a high effective rate. Besides, no recurrence was seen after an 18-year follow-up.<sup>[6]</sup> Both UNT and PWS are superficial capillaries malformation. VEGF promotes the proliferation and dilation of blood vessels, leading to the formation of PWS.<sup>[7]</sup> The immunohistochemical finding of positive staining for VEGF consolidates the participation of angiogenic factors in the pathogenesis of UNT.<sup>[8]</sup> HMME-PDT could decrease the expression of VEGF in human vascular endothelial cells, thereby reducing vascular expansion and proliferation.<sup>[7]</sup> Based on the similarities between the two diseases, we attempted HMME-PDT to treat the case and achieved excellent efficacy. But HMME-PDT also has its limitations, patients need to avoid strong sunlight exposure for 2 weeks after therapy. Second, due to the limited treatment area of HMME-PDT each time, multiple treatments may be required for UNT patients with a huge area.

In light of its effectiveness and safety, HMME-PDT may be a promising modality in the treatment of UNT, especially

for PDL-resistant cases. Nevertheless, more cases are warranted to confirm our perspective.

### Declaration of patient consent

The authors certify that they have obtained the appropriate patient consent form. In the form, the patient's parents have given their consent for images and other clinical information to be reported in the journal. The patient's parents understand that patient's name and initials will not be published and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

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### Conflicts of interest

None.

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