

**Case 15**

A 65-year-old Thai woman from Ayutthaya.

**Chief complaint:** Red spots at right chest wall for 5 years.



**Present illness:** This patient developed multiple small red spots on right anterior upper chest wall for 5 years. The lesion gradually evolved into large erythematous patches sized 7X10 centimeters. She had mild pruritus but there was no tenderness. She had ginkgo biloba and grape seed extract for food supplement.

**Past history**

She was diagnosed with diabetes mellitus, hypertension and dyslipidemia.

**Physical examination**

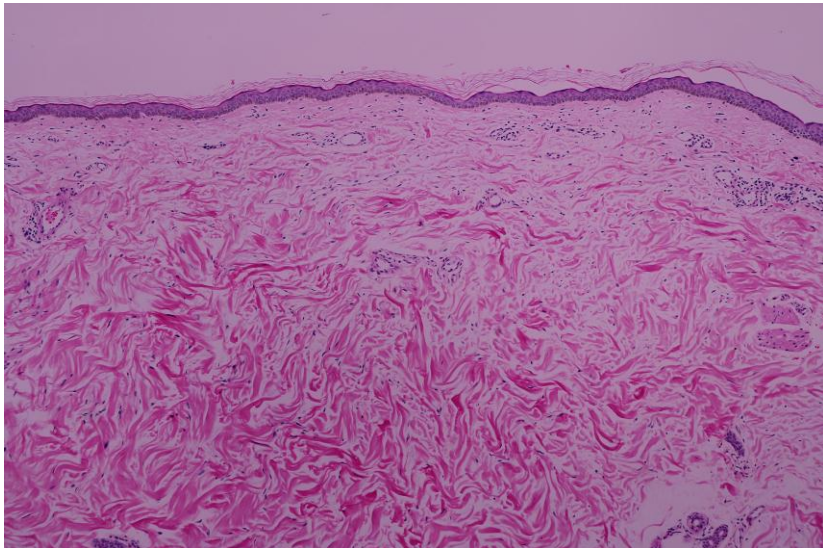
Her vital sign, HEENT, cardiovascular, respiratory and abdominal examinations were normal. No palpable lymph node.

**Skin examination**

Multiple patches of superficial telangiectasia aggregated at right anterior neck and upper chest wall.

**Histopathology** (punch biopsy from right side of chest)

- Dilated superficial blood vessels with sparse perivascular lymphocytic infiltrate in the upper dermis.

**Investigation:**

Fasting blood sugar 149 mg/dl

**Diagnosis:** Unilateral nevoid telangiectasia

**Treatment:** The patient was informed about the course of disease and choices of treatments.

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**Discussion:**

Unilateral nevoid telangiectasia (UNT) is a rare, usually acquired condition described for the first time by Blaschko in 1899<sup>1</sup>. It is a cutaneous condition consisting of congenital or acquired patches of superficial telangiectasia in a unilateral linear distribution. Although it is thought to be a rare condition with only fewer than 100 reported cases<sup>2</sup>, however, we believe it is more common as the incidence could be under reported since it is a benign condition.

UNT is considered a primary telangiectases condition. Differential diagnosis include: simple stellate angioma, Hutchinson's angioma serpiginosum, familial hemorrhagic telangiectasia, or Rendu-Osler-Weber syndrome, persisting eruptive macular telangiectasia and generalized essential telangiectasia.

The pathogenesis of UNT remains unknown. In 20% of the cases with UNT, systemic pathologies causing estrogen elevations are accused, in the other 80%, no pathologies are detected at all.<sup>3</sup> It was reported in association with puberty, pregnancy, the use of oral contraceptives and liver diseases. Why the pathology is distributed in a strict unilateral manner is unclear; however, it is thought that a localized increase in estrogen receptors caused by a chromosomal mosaicism, that is unmasked at times of relative estrogen excess, accounts for the distribution that follows Blaschko's lines.<sup>4</sup>

Clinically, UNT is characterized by multiple, unilateral blanching telangiectases arise in linear patterns or Blaschkoid pattern. It's lesion is usually asymptomatic. Predilection sites are the face, neck, shoulder-arm region, and thorax.<sup>5</sup>The third and fourth cervical dermatomes are the most common sites.<sup>6</sup>

Our case shows no sign of chronic liver disease, with normal

LFT and histologically there are sparse inflammatory cell infiltrate of lymphocytes in association with multiple dilated capillary in upper dermis.

UNT is essentially sporadic, but it has also been observed on both grandfather and grandchild.<sup>7</sup> Furthermore, some cases of UNT accompanied by Becker nevus have also been explained by mosaicism.<sup>7</sup>

UNT typically persists but rarely, in acquired cases, resolves spontaneously if the eliciting factor is removed.<sup>4</sup>

The remaining cases may be approached cosmetically with the use of camouflage, long pulse Nd YAG laser or pulse dye laser.<sup>8</sup> In this case, laser treatment was not performed since the disease represented only an aesthetic condition, which is not a cause of concern in this patient.

## References

1. Hynes LR, Shenefelt PD. Unilateral nevoid telangiectasia: occurrence in two patients with hepatitis C. *J Am Acad Dermatol.* May 1997;36(5 Pt 2):819-822.
2. Dadlani C, Kamino H, Walters RF, Rosenman K, Pomeranz MK. Unilateral nevoid telangiectasia. *Dermatol Online J.* 2008;14(10):3.
3. Smith KE, Fenske NA. Cutaneous manifestations of alcohol abuse. *J Am Acad Dermatol.* Jul 2000;43(1 Pt 1):1-16; quiz 16-18.
4. Guedes R, Leite L. Unilateral nevoid telangiectasia: a rare disease? *Indian J Dermatol.* Mar 2012;57(2):138-140.
5. Kreft B, Marsch WC, Wohlrab J. Unilateral nevoid telangiectasia syndrome. *Dermatology* 2004;209(3):215-217.
6. Selmanowitz VJ. Unilateral nevoid telangiectasia. *Ann Intern Med.* Jul 1970;73(1):87-90.
7. Happle R. Mosaicism in human skin. Understanding the patterns and mechanisms. *Arch Dermatol.* Nov 1993;129(11):1460-1470.
8. Lencastre A, Lopes MJ. Case for diagnosis. *An Bras Dermatol.* Apr 2013;88(2).