

Two Cases of Unilateral Nevoid Telangiectasia

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Unilateral nevoid telangiectasia(UNT) is a rare disorder and is characterized by superficial telangiectatic lesions distributed along the dermatomes unilaterally and usually on the upper part of the body. It has been said to be related to increased estrogen receptors in the involved skin. The lesions may be congenital or acquired. Acquired cases are typically associated with physiologic conditions such as pregnancy, puberty, hormonal therapy or cirrhosis, but the condition is also described in alcoholism without cirrhosis, carcinoma metastatic to the liver, and in hepatitis C.

We present two cases of UNT in a 26-year-old man with heavy alcoholism and in a pubertal 16-year-old girl. (*Ann Dermatol* 12(3) 185~188, 2000).

Key Ward : Unilateral nevoid telangiectasia, Alcoholism, Female puberty

Unilateral nevoid telangiectasia(UNT) was first described by Blaschko in 1899¹ and the term was first proposed by Selmanowitz in 1970². Other designations include "linear telangiectasia," "unilateral spider nevi," "unilateral telangiectasia"³ and "unilateral dermatomal superficial telangiectasia."⁶ A thorough review was presented by Wilkin et al. in 1983⁷.

The condition is characterized by superficial telangiectatic lesions distributed along the dermatomes unilaterally and usually on the upper part of the body. Histopathologically, numerous thin-walled dilated vessels are seen in the upper and middle dermis and, to a lesser extent, in the deeper part of the dermis. It has been proposed that the changes seen in this condition are induced by an increase in estrogen receptors in a dermatomal distribution⁷.

To our knowledge, three cases of UNT^{8,9} and one case of UNT associated with acanthosis nigri-

cans¹⁰ have been reported in the Korean literature. In this report, we present two additional cases of UNT in a 26-year-old man with heavy alcoholism and in a pubertal 16-year-old girl. Also, we review cases previously published in the Korean literature.

CASE REPORT

Case 1.

A 26-year-old man was seen in September 1997 for evaluation of progressive telangiectases on his left neck, shoulder and upper arm which had been present for 4 years. He had a history of heavy alcoholism for 6 years but, no history of hepatitis or medications. The family history was unremarkable.

On physical examination, there were no palmar erythema, ascites, hepatosplenomegaly, or other findings to suggest liver disease. On examination of the skin, unilaterality of involvement was most obvious on the left side of the neck, shoulder and upper arm contrasted with the clear right side (Fig. 1). The morphologic features of individual lesions consisted primarily of numerous wiry telangiectases (Fig. 2). A biopsy specimen of the telangiectases showed dilated thin-walled vessels in the upper and middle dermis (Fig. 3).

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Fig. 1. Unilateral distribution of telangiectases on the left side of the neck, shoulder and upper arm in case 1.

Fig. 3. Dilated thin-walled vessels are seen in the upper dermis in case 1 (H&E, $\times 100$).

Laboratory results revealed normal complete blood cell count and urinalysis with microscopy. Serum chemistry showed slightly elevated serum alanine aminotransferase. Hepatitis A virus anti-

Fig. 2. Closed-up of case 1 lesions shows that the lesions consist primarily of numerous wiry telangiectases.

Fig. 4. Numerous wiry telangiectases on the extensor area of the left arm in case 2.

body (IgM), hepatitis B virus surface antigen/antibody and Hepatitis C virus antibody were negative. A chest x-ray film showed normal findings and an ultrasonography of the liver and gallbladder showed normal findings. Gastrofibroscopy showed mild erosive gastritis.

Case 2.

A 16-year-old girl was seen in June 1997 for evaluation of progressive telangiectases on the extensor area of the left arm which had been present for 3 years. There was no history of hepatitis or medications. The family history was unremarkable.

On physical examination, there were no palmar erythema, ascites, hepatosplenomegaly, or other findings to suggest liver disease. Skin lesions showed numerous wiry telangiectases on the ex-

Table 1. Summary of unilateral nevoid telangiectasia cases in the Korean literature

No.	Authors	Sex/Age	Underlying condition	Dermatomal distribution
1	Ahn CH et al, 1981 ⁸	M/17	Puberty	Left C3-5, T1-2
2	Jang IS et al, 1983 ⁹	M/23	Puberty	Right T2-4, L5, S1-2
3	Jang IS et al, 1983 ⁹	M/25	Puberty	Left C2-5, T1
4	Park SM et al, 1995 ¹⁰	M/20	Puberty	Right C5-6
5	Present study	M/26	Heavy alcoholism	Left C3-7, T1-2
6	Present study	F/16	Puberty	Left C5-7

tensor area of the left arm (Fig. 4). A biopsy specimen of the telangiectases showed dilated thin-walled vessels in the upper and middle dermis as in case 1.

Laboratory results revealed normal complete blood cell count, erythrocyte sedimentation rate, urinalysis with microscopy and serum chemistry. A chest x-ray film showed normal findings.

DISCUSSION

Most cases occur in women during pregnancy or puberty and are localized to the upper body⁷. In men alcoholic cirrhosis is the most common association⁷. Also, it has been expressed congenitally^{2,3,8,7,11,12}, during male puberty^{13,14}, and with carcinoid syndrome with liver metastasis¹⁵. There were also reports of the condition arising in a male alcoholic without cirrhosis¹⁶ and in two males with hepatitis C¹⁷. Thus, we suspect that our patient in case 1 might result from heavy alcoholism without evidence of cirrhosis, hepatitis C or carcinoid syndrome with liver metastasis and the patient in case 2 from female puberty.

It has been proposed that the changes seen in this condition are induced by an increase in estrogen receptors in a dermatomal distribution⁷. But, the commonly accepted theory that elevated circulating estrogen acts as a stimulus for the development of this disorder has been questioned because of the normal circulating estrogen level¹⁸. Uhlin and McCarty in 1983 demonstrated an increase in both estrogen and progesterone receptors in the involved skin with a method that has been effective in evaluating the low levels of estrogen receptors in the skin^{19,20}. Recently, Hynes and Shenefelt in 1997 proposed that UNT might result from a localized increase in estrogen levels caused by a chromosomal mosaicism that was unmasked at times of relative es-

trogen excess¹⁷. They believed that this mechanism could account for the distribution that followed Blaschko's lines. However, we suspect that the lesions in our cases were induced by the increased circulating estrogen level because heavy alcoholism or female puberty might be associated with that.

Histopathologically, numerous thin-walled dilated vessels are seen in the upper and middle dermis and, to a lesser extent, in the deeper part of the dermis⁷. Based on clinical and histopathological findings, our cases are consistent with UNT.

In conclusion, three cases of UNT^{8,9} and one case of UNT associated with acanthosis nigricans¹⁰ have been reported in the Korean literature (Table 1). But, there was no report associated with heavy alcoholism or female puberty. We describe two cases of UNT associated with heavy alcoholism and female puberty.

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