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CASES FOR DIAGNOSIS

Angiomatous Rash in a Pregnant Woman

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Clinical History

The patient was a 27-year-old woman of 5 months gestation who was seen for a rash on the trunk that had developed towards the end of the first trimester of pregnancy. The relevant findings in her past medical history were that she was a hepatitis C virus carrier and had had an uncomplicated previous pregnancy and a voluntary abortion. She had not received any treatment in the previous months except for vitamin supplements and did not report easy bleeding or a personal or family history of coagulation disorders.

Physical Examination

Examination revealed the presence of small, red-violaceous, spidery maculopapular lesions of vascular appearance with a unilateral distribution affecting the scapular region, shoulder, breast, and posterior aspect of the arm on the right side. The lesions disappeared under pressure (Figures 1 and 2).

Complementary Tests

Laboratory studies including complete blood count, biochemistry, and serology for syphilis, hepatitis B virus, and human immunodeficiency virus were normal. The only significant findings were the presence of antibodies to hepatitis C virus (already known) and high levels of estrogens (consistent with her gestational state).

Histopathology

Biopsy of a lesion was performed and histological examination revealed the presence of a normal epidermis with dilated capillaries in the superficial dermis (Figure 3).

What Was the Diagnosis?

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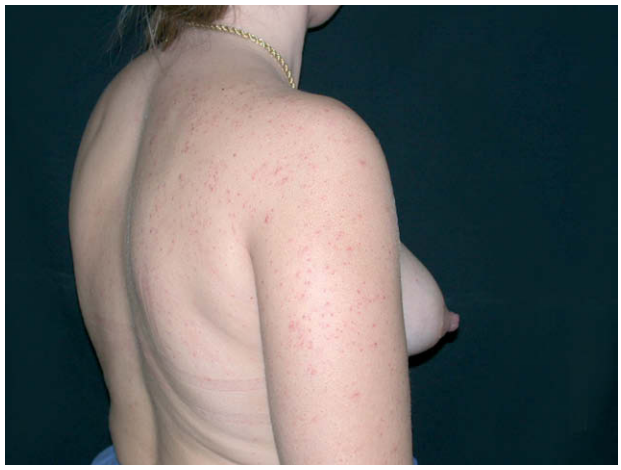

Figure 1.

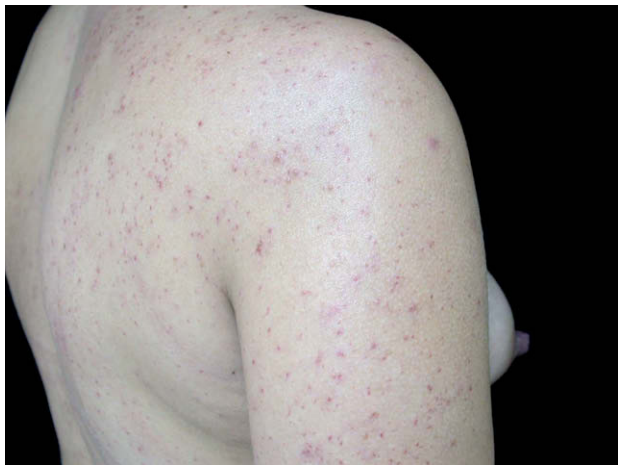

Figure 2.

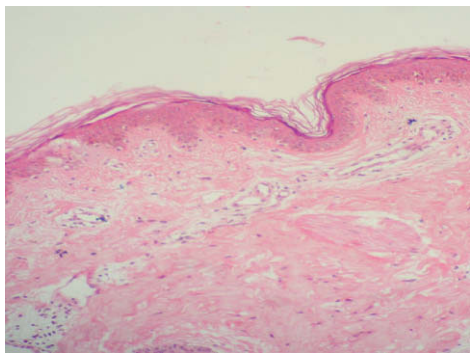

Figure 3. Hematoxylin-eosin, ×40

Diagnosis

Unilateral nevoid telangiectasia.

Clinical Course and Treatment

In view of the benign nature of this disorder, it was decided to maintain the patient under observation. Six months after giving birth, the lesions had disappeared.

Discussion

In the early 20th century, Blaschko and Zeisler were the first to describe the appearance of acquired telangiectasias with a nevoid distribution. Later, in 1970, Selmanowitz proposed the term unilateral nevoid telangiectasia. Few cases have been reported in the literature since that time and today it is a relatively unknown disorder.

Unilateral nevoid telangiectasia syndrome is characterized by the appearance of unilateral telangiectasias with a segmental distribution. In the majority of cases the lesions are situated in the trigeminal area and between dermatomes C3 and T1. Apart from the skin, they can also affect the oral and gastric mucosas. The right side appears to be affected more frequently than the left, and it is most common in young women. In 1977, Wilkin[1] performed a review of this disorder and classified it into 2 categories: congenital (with very few cases described) and acquired. The acquired forms may be secondary to states of hyperestrogenism (pregnancy,[2] puberty, and menstrual cycle); they have also been reported in association with liver disease (alcoholic, viral, metastatic). In general, when telangiectasias appear due to changes in estrogen levels, there are usually no systemic manifestations; in contrast, when they are associated with liver disease, gastric lesions may be found.

The etiology is unclear, and a number of theories have been proposed. There is speculation on the role of the sex hormones (estrogens and progestogens) as a trigger for the disorder,[3] changes in the concentration of adrenergic receptors, which could contribute directly or indirectly to capillary dilatation, or an increase in estrogen receptors. Additionally, certain prostaglandins[3] and epidermal angiogenic factors[4] (vascular endothelial growth factor, epidermal growth factor) may also be involved in the pathogenesis.

The segmental and unilateral localization of the telangiectasias may be due to an abnormal distribution of cells sensitive to estrogens during the embryonic period.[1] The lesions occasionally follow a pattern along the Blaschko lines, an observation which would be consistent with mosaicism.[5]

The diagnosis of unilateral nevoid telangiectasia syndrome is clinical and is based on the appearance of telangiectasias with a unilateral and metameric distribution (particularly affecting the dermatomes in the cervical and trigeminal regions).[6]

The differential diagnosis principally involves other disorders in which telangiectasias are a feature, such as angioma serpiginosum, hereditary hemorrhagic telangiectasia, simple stellate angioma or spider angioma, telangiectasia macularis eruptiva perstans, and generalized essential telangiectasia.

The lesions tend to resolve once the hormone levels have normalized.

We present a case of unilateral nevoid telangiectasia in a pregnant woman who was a hepatitis C virus carrier. We believe that these 2 factors may have an additive effect that could trigger the syndrome.

Conflicts of Interest
The authors declare no conflicts of interest.

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