Unilateral Nevoid Telangiectasis in a Patient With Chronic Hepatitis B Virus Infection

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To the Editor:
Unilateral nevoid telangiectasis syndrome consists of multiple telangiectasias with a metamer distribution and is considered an essential telangiectasia. Different theories have been suggested regarding its pathogenesis, but the most widely accepted is related to variations in estrogen levels or their receptors. We present a new case of this syndrome, possibly associated with chronic hepatitis B virus (HBV) infection.

The patient was a 47-year-old man who was referred due to asymptomatic erythematous lesions of 2 months duration, located on the right arm, neck, and upper part of the trunk. Of note, he had a personal history of obstructive sleep apnea syndrome and was a former smoker. Physical examination showed multiple vascular lesions consisting of telangiectasias with a metamer distribution in the upper third of the trunk, neck, and dorsal and external surfaces of the right arm (Figure 1 and Figure 2). Blood analysis demonstrated high cholesterol and triglyceride concentrations, without other findings of note (luteinizing hormone [LH], follicle-stimulating hormone [FSH], dehydroepiandrosterone, sex hormones, or glutamic pyruvic transaminase and glutamic oxalacetic transaminase). However, serological tests were positive for hepatitis B surface antigen (HBsAg), HBV core antibodies and HBVe antibodies, with no detectable viral load, which is consistent with the nonreplicative phase of chronic HBV infection, indicating that the carrier was healthy. There was no evidence of liver cirrhosis.

Figure 1. Posterior view showing the metamer distribution of the telangiectasias.

Figure 2. Anterior view showing how the telangiectasias do not cross the midline.
After punch biopsy, a histological analysis was conducted for the differential diagnosis of other diseases that might cause telangiectasias. Vascular dilatation was found but there were no other significant abnormalities.

Telangiectasias are thin dilated blood vessels near the surface of the skin that can be seen through the skin and mucous membranes. They can present as isolated lesions or form part of specific clinical entities. They are classified into 2 groups: essential, when the origin is unknown, and secondary, when they are caused by other dermatological or systemic abnormalities. The essential form may appear at birth, during infancy, or even in adults.

Unilateral nevoid telangiectasis syndrome is classified within the group of essential telangiectasias. It was first described by Blaschko in 1899,1 and in 1970 Selanowitz2 proposed the term unilateral nevoid telangiectasia, but it was not until 1977 that Wilkin3 classified it into a congenital form and an acquired form (the most frequent).

Most cases described appear in situations where there is an increase in estrogen levels, such as pregnancy,4 hormone replacement therapy and contraception, other drugs, puberty, and liver cirrhosis. Cases associated with ovarian carcinoid tumors and serological tests positive for hepatitis C virus (HCV) have also been reported.5 Thus, some authors recommend administering a battery of complementary tests in these patients which should include the following: pregnancy test, hormonal study (androgens, estrogens, LH/FSH, thyroid, etc), liver function test, and serologic tests for liver-tropic viruses.

Its pathogenesis remains uncertain, and has been mainly associated with increased serum estrogen levels or an increase in the number or sensitivity of estrogen or progestogen receptors with a metameric distribution, thus these receptors would be active in these locations.6 The most widely accepted basis for these abnormalities is mosaicism among these patients, that is, a somatic mutation during embryological development. The affected areas would therefore be more susceptible to possible hormonal variations, as occurs in physiological situations (puberty, pregnancy, etc) or pathological situations (alcoholism, tumors, hepatitis due to HCV, or HBV as in our patient, etc). The fact that this condition has been linked, in some cases, with other lesions—for example, inflammatory linear verrucous epidermal nevus, which has a similar pathogenesis—lends weight to the mosaicism hypothesis.

This hypothesis is also shared by Karakas et al.,7 who reported a case of unilateral nevoid telangiectasia associated with HBV infection. A woman presented with involvement of the face and neck, with no analytical or histological abnormalities, but serological tests showed that she was an HBsAg carrier. Unlike the present case, no other serological markers of HBV infection were found.

Clinical characteristics include fine telangiectasias and spider veins with a metameric distribution and that most frequently affect the C3-T1 dermatomes; however, some authors argue that they follow the lines of Blaschko, and may not disappear under finger or diascopic pressure.8

The diagnosis is basically clinical; anatomicopathological study shows capillary vasodilatation with mild lymphohistiocytic infiltrate around the vessels and their subsidiaries.

Generally, the lesions are persistent, although spontaneous resolution postpartum has been described. Treatment may not be definitive, and mainly consists of radiosurgery or laser surgery (especially pulsed dye laser), although this disease has a benign prognosis.

We present a new case of unilateral nevoid telangiectasia, with the unusual characteristic of the patient being a healthy carrier of HBV.