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An Unusual Manifestation of Linear Atrophoderma of Moulin

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Sir,

Moulin et al. (1) first described linear atrophoderma in 1992 and differentiated this uncommon condition from other linear dermatoses. Clinically, it is present as pigmented and atrophic bands or lines that follow Blaschko's lines exactly, with no preceding inflammation or subsequent induration or scleroderma.

To date, 11 cases of linear atrophoderma of Moulin (LAM) have been reported in the literature (1–7). Here, we present a woman with two types of skin lesions: linear atrophic lesions on the right arm and on the left mandibular region along with atrophic areas and telangiectatic macules in linear arrangement following the course of Blaschko's lines on the right buttock and leg.

The expression of the disease in our case does not conform exactly to any of the reported cases with classic cutaneous manifestations characteristic for LAM.

CASE REPORT

A 20-year-old woman presented with a 4-year history of asymptomatic, unilateral linear skin lesions following the appearance of Blaschko's lines on the arms and legs and on the face. Her medical and family history was unremarkable. The disease had a chronic course without progression or regression.

Physical examination revealed two types of skin lesions: hypopigmented, atrophic, slightly depressed linear lesions on the medial aspect of the right arm, from the axilla to the wrist and on the left mandibular region, and 1-cm wide atrophic band-like lesions along with multiple telangiectatic macules following Blaschko's lines on the right buttock and leg (Fig. 1).



Fig. 1. Linear atrophic telangiectatic lesions on the right buttock and leg. The pattern is consistent with Blaschko's lines.

Laboratory findings, including a complete blood count, urinalysis and liver function test, were normal. Complete ophthalmologic, odontologic and radiologic surveys revealed no abnormalities.

A skin biopsy taken from the atrophic lesion with telangiectasias on the right leg revealed moderate psoriasiform epidermal hyperplasia with hyaline eosinophilic bodies in the spinous layer; oedema and dilated small blood vessels in the papillary dermis, some with hyalinization; scant inflammatory infiltrate of lymphocytes and plasma cells and some hyalinization of the collagen in the upper dermis. The elastic tissue is diminished (Fig. 2). These histologic features were considered to be compatible with the diagnosis of LAM.

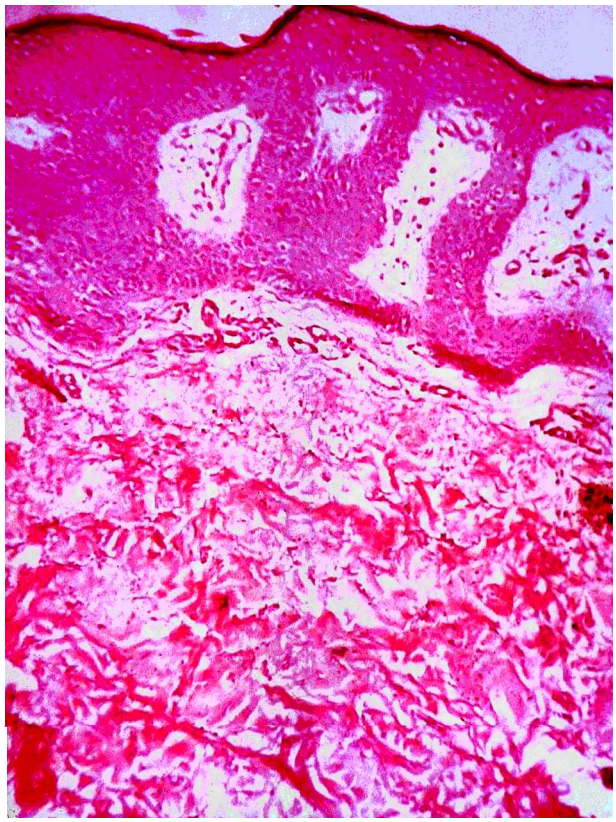


Fig. 2. Moderate psoriasiform epidermal hyperplasia, scanty inflammatory infiltrate, oedema and dilated blood vessels.

DISCUSSION

The diagnosis of LAM is usually established on the basis of the classic cutaneous findings: pigmented atrophic bands or lines localized mostly on the trunk, following Blaschko's lines, without preceding inflammation or subsequent induration or sclerosis. The lesions appear during childhood or adolescence, between the ages of 6 and 20, and are always unilateral (1, 6, 7). The intensity of pigmentation and atrophy is variable (1). Browne & Fisher (6) noted a preceding inflammatory phase both clinically and histologically and proposed that LAM had an antecedent inflammatory phase that ultimately evolved into hyperpigmentation with atrophy. They suggested two variants of LAM: an inflammatory and a non-inflammatory. This would be analogous to anetoderma, in which there is an inflammatory Jadassohn-Pellizzary subtype and a non-inflammatory Schweninger-Buzzi subtype.

Our patient showed many findings similar to those of previously described LAM cases. The lesions first appeared at the age of 17 and after a short period of rapid enlargement the skin disorder remained unchanged. Skin lesions were asymptomatic and unilateral, localized on the limbs and on the mandibular region. They showed no signs of inflammation, induration or sclerosis. We observed an association of hypopigmented atrophic lesions and telangiectatic macules following the course of Blaschko's lines. Although in LAM the intensity of pigmentation may be variable, pigmentary changes were not present in our case. An additional clinical feature observed in our patient is telangietasias, which has never been reported in any cases with LAM so far.

Our patient's disease does not precisely resemble any of the large variety of conditions following Blaschko's lines, e.g. incontinentia pigmenti, hypomelanosis of Ito, linear and whorled nevoid hypermelanosis, lichen striatus, lichen nitidus or scleroderma (8). The skin lesions of atrophoderma of Pasini and Pierini resemble those of LAM; however, they do not follow Blaschko's lines (6, 7). Linear scleroderma can be regarded as an important differential diagnosis of LAM. Linear scleroderma shows similar configuration, atrophy, hyperpigmentation and may include telangiectasia formation. The absence of inflammation, sclerosis or induration led to a diagnosis of LAM (9, 10). In our case, focal dermal hypoplasia (FDH) should be included in the differential diagnosis. The abnormalities seen in this multisystem disease include atrophic skin lesions, linear and reticular areas of telangiectasias, hypopigmentations, hyperpigmentations and papillomas, as well as skeletal, dental and ocular defects (11). The clinical features of FDH show a wide range of variability. Several cases of FDH with minimal cutaneous and extracutaneous manifestations have been described (12). Other patients with FDH and clinical findings limited entirely to the skin have also been reported (13).

In conclusion, the onset, the course and the clinical and histologic findings in our case are more consistent with a diagnosis of LAM than of linear scleroderma, FDH or other Blaschko linear dermatoses. A non-inflammatory variant of LAM is probably heterogeneous. We support the concept that our case may be an unreported form of LAM associating atrophic hypopigmented macules with a prominent telangiectatic component.

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