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A Case of Linear Atrophoderma of Moulin

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To the Editor:

Linear atrophoderma of Moulin is characterized by slightly atrophic hyperpigmented patches that follow Blaschko lines. Only a few cases have been reported since the condition was first described by Moulin et al[1] in 1992 and most of these have been isolated cases. Moreover, not all of them coincide with the original description. We report the case of a patient with typical clinical and histologic findings.

A 17-year-old male patient presented with hyperpigmented lesions on the right upper arm. The lesions, which had appeared 12 months earlier, occurred as multiple brown macules that formed a distinctive S-shaped curve along the affected arm. Since their onset, they had spread slowly and progressively, grown in number and size, darkened, and acquired a slightly atrophic texture (Figure 1). There were no subjective or objective symptoms, related events, or inflammatory reactions in the affected area. The 2 skin biopsies performed revealed only localized hyperpigmentation in the basal layer of the epidermis (Figure 2). The results of the other tests performed (complete blood count, coagulation, liver and kidney function, antinuclear antibodies, protein profile, erythrocyte sedimentation rate, chest radiograph, and serological tests for *Borrelia*) were all normal. No specific treatment was prescribed and, with the exception of the darkening of the atrophic patches, the condition remained unchanged during the first 6 months of follow-up. Four years later, the lesions seem to be stable and there have been no evident changes.

Linear atrophoderma of Moulin is a rare skin condition featuring lesions that follow Blaschko lines.[1,2] In our review of the literature, we found 22 publications describing the condition (Table). Because several of the clinical and histologic features described do not adhere strictly to the original description provided by Moulin et al,[1] the true number of cases may actually be smaller.

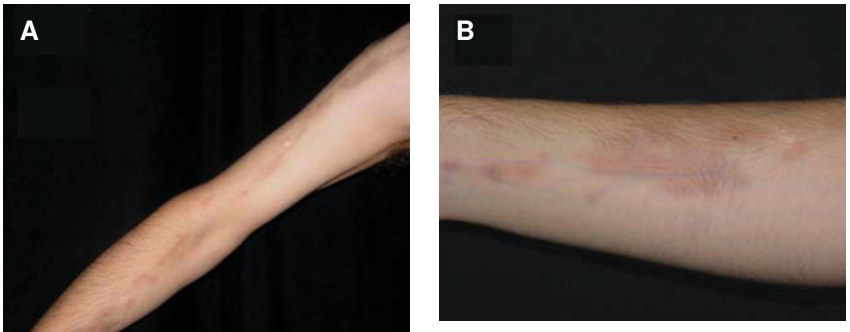

Figure 1. A, Multiple brown macules following Blaschko lines on upper arm. B, Close-up of the slightly atrophic appearance of the hyperpigmented lesions.

Table. Cases Reported as Linear Atrophoderma of Moulin

	Year	*No. of Patients*	*Sex*	*Age at Onset*	*Lesion Site*	*Histologic Findings*
Moulin et al[1]	1992	1	M	8	Left part of trunk	Hyperpigmentation of basal epidermis
		2	F	7	Right part of trunk	Hyperpigmentation of basal epidermis
		3	M	15	Right part of trunk	Hyperpigmentation of basal epidermis
		4	M	20	Left part of trunk	Biopsy not performed
		5	M	6	Left part of trunk and left arm	Biopsy not performed
Baumann et al[2]	1994	6	M	22	Right part of trunk and right arm	Ballooning in basal epidermis, perivascular lymphocytic infiltrate, and increased collagen in the dermis.
Larregue et al[9]	1995	7	M	15	Left part of trunk	Increased collagen in dermis
Wollenberg et al	1996	8	F	11	Right arm	Epidermal atrophy, perivascular lymphocytic infiltrate, and increased collagen in the dermis.
Artola et al[8]	1996	9	F	9	Left part of trunk	Acanthosis and hyperpigmentation of basal epidermis, perivascular lymphocytic infiltrate, and increased collagen in the dermis.
Cecchi et al[4]	1997	10	F	12	Right part of back and right arm	Localized hyperpigmentation of basal epidermis
Browne et al5	2000	11	M	13	Limbs and trunk, bilateral	Acanthosis, hypogranulosis, and parakeratosis with perivascular lymphocytic infiltrate in the dermis
Rompel et al[10]	2000	12	F	14	Right part of trunk and right buttock	Hyperpigmentation in basal epidermis, Civatte bodies, perivascular lymphocytic infiltrate, and increased collagen.
Martin et al[14]	2002	13	M	9	Left part of trunk	Perivascular lymphocytic infiltrate and increased collagen
Miteva et al[13]	2002	14	F	16	Right part of face, right arm and leg	Psoriasiform epidermal hyperplasia, perivascular lymphocytic infiltrate, and increased collagen in the dermis.
Danarti et al[3]	2003	15	F	14	Left part of trunk and left arm	Perivascular lymphocytic infiltrate
		16	F	24	Left part of trunk and left arm	Biopsy not performed
		17	F	38	Left thigh	Unremarkable epidermis and dermis
		18	F	15	Left buttock and left iliac crest	Biopsy not performed
Utikal et al[6]	2003	19	M	23	Limbs and trunk, bilateral involvement	Perivascular lymphocytic infiltrate and edema in dermis
		20	F	2		
Miteva et al[12]	2005	21	M	9	Left part of trunk and left arm	Hyperkeratosis, irregular acanthosis, hyperpigmentation of basal epidermis, and increased collagen in dermis
Atasoy et al[7]	2006	22	M	16	Right part of trunk and right arm	Epidermal atrophy, perivascular lymphocytic infiltrate, and fragmented collagen fibers
Present case	2007	23	M	16	Right upper arm	Localized hyperpigmentation of basal epidermis

Abbreviations: F, female; M, male.

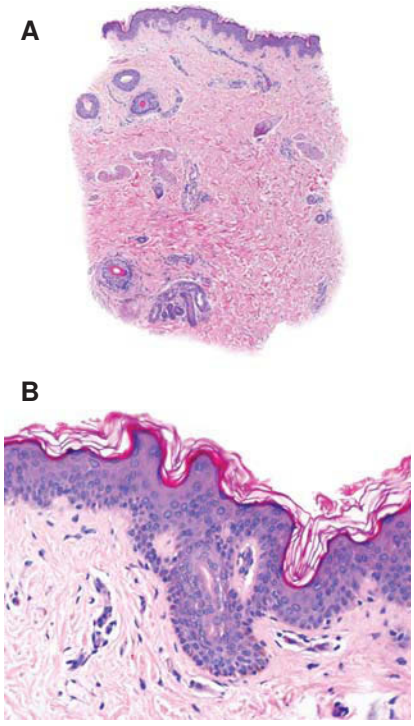

Figure 2. A, Unremarkable epidermal and dermis. B, Focal hyperpigmentation of basal membrane (Hematoxylin–eosin).

Strictly speaking, the diagnostic criteria for linear atrophoderma of Moulin include (1) onset during childhood or adolescence; (2) development of hyperpigmented, slightly atrophic, unilateral lesions following Blaschko lines on the trunk or limbs; (3) absence of prior inflammation or subsequent scleroderma; (4) a stable, nonprogressive clinical course without a pattern of remission; and (5) histologic findings showing hyperpigmentation of the basal epidermis and a normal dermis with unaltered connective tissue and elastic fibers.[1-4]

One study reported symmetric lesions on the lower limbs; these lesions gradually extended upwards and were accompanied by telangiectasia.[5] Prior inflammation has been reported in patients with this variant.[5,6]

As suggested by Utikal et al,[6] it would seem more logical to assume that the conditions described were separate clinical entities rather than atypical variants of linear atrophoderma of Moulin, which is in itself a rare condition. Perhaps the authors were describing childhood-onset cases of linear nevoid atrophoderma with telangiectasias in patients without an associated hormonal disorder. Atasoy et al[7] recently observed leuconychia in a patient with symptoms consistent with linear atrophoderma of Moulin.

The main differences between the cases reported to date are related to histologic findings. The most common finding is a perivascular lymphocytic inflammatory infiltrate in the superficial dermis combined with abnormal collagen fibers.[2,3,5-10] Because perivascular lymphocytic infiltrates and abnormal collagen fibers are more characteristic of atrophoderma of Pasini and Pierini than of linear atrophoderma of Moulin, Ang et al[11] proposed naming this condition Blaschko-linear atrophoderma of Pasini and Pierini. Histologic changes in the epidermis—in particular atrophy,[6,7] acanthosis,[5,8,12] and hypogranulosis, parakeratosis, and hyperkeratosis[5,10,13,14]—have also been reported, although not as frequently. It is very probable that those cases were actually describing epidermal nevi, linear inflammatory epidermal nevi, lichen striatus, or nevoid hypermelanosis.[12,13]

In conclusion, based on the clinical and histologic findings documented by several authors describing what they considered to be linear atrophoderma of Moulin, we believe that the prevalence of the condition may be overestimated as several of these authors reported histologic findings that are compatible with other clinical entities.

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