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Linear and Annular Lupus Panniculitis of the Scalp[☆]

Paniculitis lúpica de distribución lineal y anular localizada en el cuero cabelludo

To the Editor:

Lupus panniculitis (LP) is a rare variant of cutaneous lupus erythematosus (CLE) that primarily affects the deep dermis and adipose tissue.¹ It develops in 1% to 3% of patients with CLE. Antinuclear antibodies (ANA) are positive in 70% of cases, but only 25% to 50% satisfy the criteria for systemic lupus erythematosus.² The diagnosis of LP is based on the correlation of clinical, serological, and, most importantly, histopathological findings. Direct immunofluorescence is positive in 70% to 80% of cases and can help to confirm the diagnosis.² LP with a linear or annular morphology is extremely rare; only 14 cases of linear LP have been described,^{3–7} and just 1 case of LP with annular lesions⁸ (Table 1). Of these 15 cases, 9 affected the scalp.

We present the case of a 34-year-old man who consulted because of a plaque of alopecia that had been present in the right parietal region for 12 months. Three months earlier he had been diagnosed with alopecia areata but the plaque did not respond to treatment with topical corticosteroids and subsequently had increased in size and had become erythematous and painful. The patient did not report fever, joint pain, oral aphthous ulcers, or other associated symptoms. Physical examination revealed an erythematous, edematous plaque of alopecia in the right parietal region. The plaque had an annular morphology and measured about 6 cm in diameter (Fig. 1A). Skin biopsy findings were compatible with lupus panniculitis, and direct immunofluorescence revealed granular deposits of immunoglobulin M in the basement membrane. Complete blood count and serum

biochemistry were normal, and immunological study was positive for ANA at a titer of 1:320 and for Anti-Ro.

A new plaque of alopecia simulating alopecia areata was observed in the occipital region at subsequent follow-up visits (Fig. 1B).

A diagnosis of LP was made based on the clinical, histological, and serological findings, and a 5-month course of treatment with hydroxychloroquine was started at a dose of 400 mg/d. This regimen produced a reduction in the erythema and induration of the plaque and resolution of the alopecia. The patient did not attend the scheduled follow-up, but came to an unscheduled visit at 12 months, at which time he presented multiple firm erythematous nodules and nonscarring alopecia with an annular and linear morphology located in the occipital and both parietal regions (Fig. 1C). Oral prednisone was prescribed at a dose of 30 mg/d tapered over 3 weeks; there was an initial improvement, but the lesions recurred after treatment was discontinued.

Linear LP localized to the scalp was first described by Nagai et al.⁴ in 2003. A further 7 cases of LP in a linear pattern on the scalp have been described and 1 case of LP at the same site with an annular pattern.^{5–8} Most of the cases have been reported in southwest Asia,⁶ suggesting a genetic or ethnic predisposition; the condition affects both sexes and occurs mainly in young patients. Clinical presentation takes the form of plaques of alopecia; these are usually nonscarring and follow the Blaschko lines,⁶ which, on the scalp, have a spiral distribution centered on the vertex⁹ (Fig. 1D). It would appear that inflammatory dermatoses that follow the Blaschko lines are the result of a genetic mosaicism, in which a clone of abnormal cells remains inactive until an environmental factor stimulates its growth.¹⁰ The overlying skin may be normal, erythematous, or show signs of discoid lupus. The histopathology findings in linear LP include a predominantly lobular lymphocytic panniculitis with hyaline fat necrosis,⁷ as observed in other forms of LP. Our patient underwent 2 skin biopsies. The first showed a dense lymphocytic infiltrate mainly affecting the deep dermis and adipose tissue, with areas of fat necrosis (Fig. 2A) and vacuolar damage of the basal layer of the epidermis (Fig. 2B). The second biopsy revealed

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Table 1 Review of the Published Cases of Lupus Panniculitis With a Linear or Annular Pattern.

Reference	Age, y/Sex/Country	Site	Lesion morphology	Immunological Study	Treatment	Response
Tada et al., 1991	9/M/Japan	Left leg	Linear	Negative	TC	Improvement
Innocenzi et al., 1997	17/M/Italy	Breast, right arm	Linear	ANA 1:40	Not mentioned	
Tamada et al. ³ ; 1999	19/M/Japan	Left arm	Linear	Negative	IC	Improvement
Nagai et al. ⁴ ; 2003	10/F/Japan	Scalp, forehead	Linear	ANA 1:320	TC	Improvement, SA
Wu et al., 2004	21/F/Taiwan	Scalp	Linear	Negative	HCQ	CR
[1,0]Marzano et al., 2005	21/F/Italy	Scalp, forehead	Linear	ANA 1:280	OC, Cy, IVC	SLE, SA, skin atrophy
	32/F/Italy	Right arm	Linear	ANA 1:640	OC, HCQ	Skin atrophy
Bacanli et al. ⁸ ; 2005	16/F/Turkey	Scalp, axilla	Annular, linear	ANA 1:320	HCQ	Improvement, SA
Shin et al. ⁵ ; 2007	20/M/Korea	Scalp	Linear	Negative	TC, IC, OC	Improvement, skin atrophy
[1,0]Rhee et al., 2009	14/M/Korea	Scalp	Linear	Not mentioned	HCQ, OC, IC	CR
	21/M/Korea	Scalp	Linear	Not mentioned	DP	CR
Tamiya et al., 2010	6/F/Japan	Face	Linear	Negative	OC, TC	CR
Lee et al., 2011	28/F/Korea	Back, right thigh	Linear	ANA 1:2560	HCQ	Lipoatrophy
Chen et al. ⁶ ; 2012	32/M/Taiwan	Scalp	Linear	Negative	HCQ, IC	CR
Tsuzaka et al. ⁷ ; 2012	26/F/Japan	Scalp, forehead	Linear	ANA 1:1280, Anti-Ro+	OC	CR
Present case	34/M/Spain	Scalp	Annular, linear	ANA 1:320, Anti-Ro+	HCQ, OC	Improvement

Abbreviations: ANA, antinuclear antibodies; CR, complete remission; Cy, cyclophosphamide; DP, dapsone; F, female; HCQ, hydroxychloroquine; IC, intralesional corticosteroids; IVC, intravenous corticosteroids; M, male; OC, oral corticosteroids; SA, scarring alopecia; SLE, systemic lupus erythematosus; TC, topical corticosteroids.

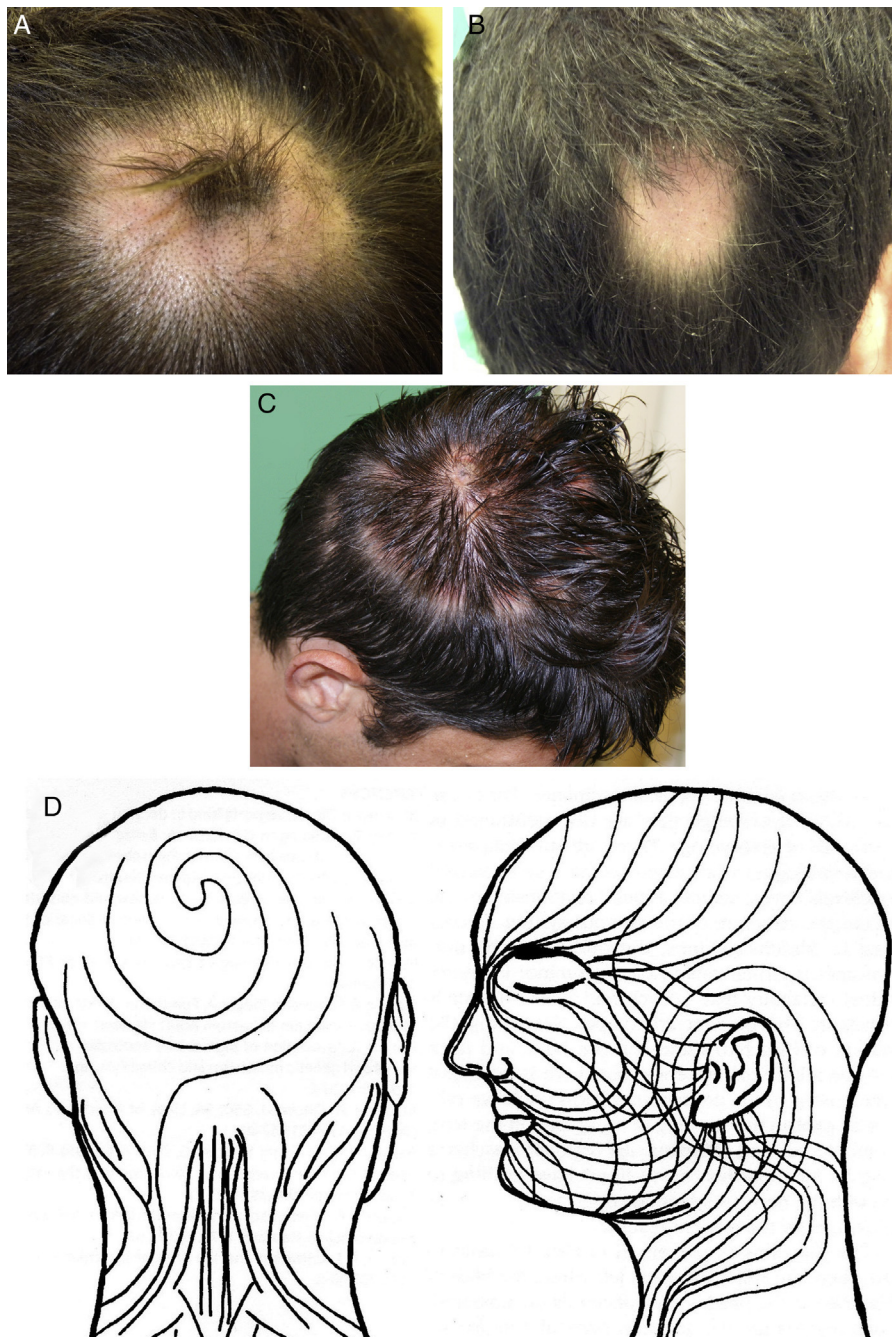


Figure 1 A, Annular plaque of alopecia situated at the vertex. B, Plaque of alopecia that simulates alopecia areata. C, Arcuate plaque of alopecia that appears to follow the Blaschko lines. D, Distribution of the Blaschko lines on the scalp (pattern proposed by Happle⁹).

nodular lymphoid aggregates in which plasma cells were observed.

The main condition to be excluded in the differential diagnosis of linear LP affecting the scalp is alopecia areata. The presence of erythema, edema, and pain supports a diagnosis of LP, although the overlying skin may show no changes in the early stages. Other conditions to be considered in the differential diagnosis are morphea en coup de sabre, lipedematous alopecia, and panniculitis-like T-cell lymphoma. The

most widely used drugs have been the antimalarials and topical, intralesional, and oral corticosteroids; these treatments have achieved a good response in most cases.⁷ In summary, linear LP is a rare clinical variant of LP distributed along the Blaschko lines that presents a linear, annular, or arcuate pattern depending on the site of the lesions.

We have reported the second case of annular LP localized to the scalp. The condition presented clinically as painful erythematous plaques of nonscarring alopecia in a

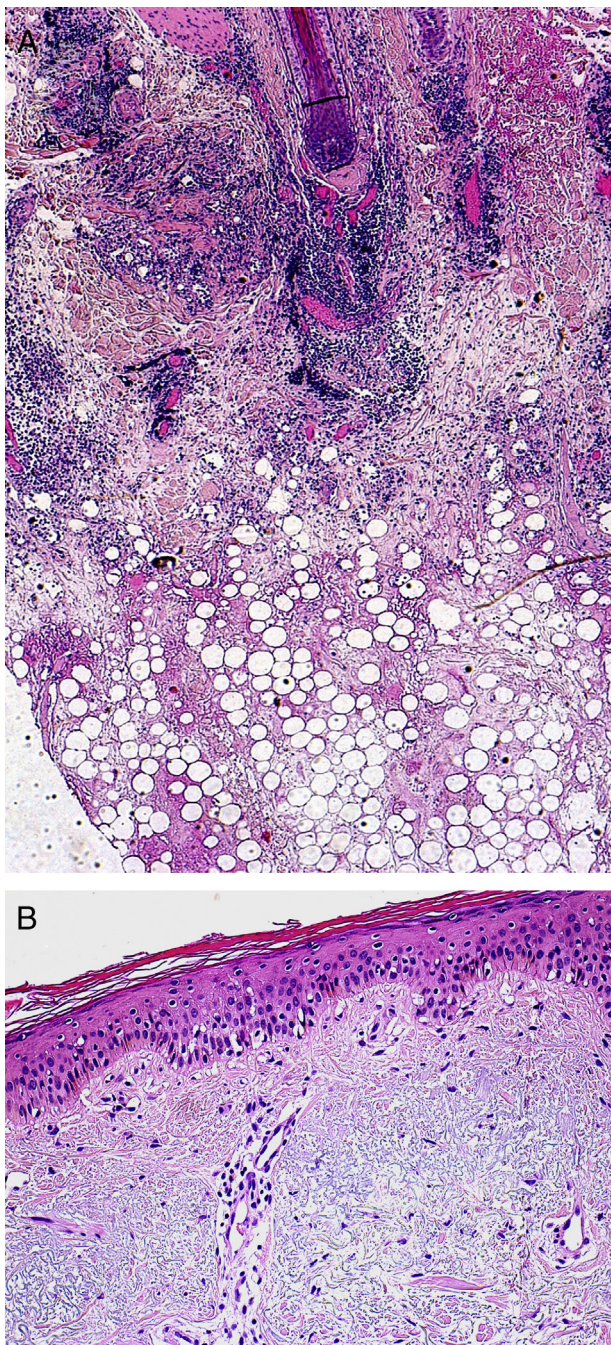


Figure 2 A, Predominantly lymphocytic infiltrate in the deep dermis and adipose tissue, with hyaline fat necrosis (hematoxylin-eosin, original magnification $\times 40$). B, Vacuolar changes in the basal layer of the epidermis (hematoxylin-eosin, original magnification $\times 200$).

young white man. LP should be included in the differential diagnosis of focal, linear, or annular plaques of alopecia situated on the scalp, particularly if they are erythematous or painful.

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