

CASE REPORT

A Case of Unilateral Ashy Dermatosi

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Ashy dermatosis, also known as erythema dyschromicum perstans, is a peculiar, slowly progressive, idiopathic dermal melanosis. In most cases, slate gray- to lead-colored patches are symmetrically distributed over the body. Ashy dermatosis with a unilateral distribution is rare. We report a case of unilateral ashy dermatosis in a 27-year-old Korean man. (*Ann Dermatol* 21(4) 432~434, 2009)

-Keywords-

Ashy dermatosis, Erythema dyschromicum perstans, Unilateral

INTRODUCTION

Ashy dermatosis is a peculiar, slowly progressive, idiopathic dermal melanosis that was first described by Ramirez in 1957¹. Clinically, ashy dermatosis consists of slate gray- to lead-colored patches and ranging in size from 3 mm to very large confluent patches, surrounded by an erythematous, slightly elevated border^{2,3}. In most cases, the trunk, arms, neck, and face are involved symmetrically². Only three cases of unilateral ashy dermatosis have been reported (Table 1)⁴⁻⁶.

CASE REPORT

A 27-year-old Korean man presented to our clinic with progressive cutaneous pigmentary changes on his left back, flank, and abdomen. One year earlier, erythematous

Table 1. Reported cases of unilateral ashy dermatosis

Author (Year)	Age/Sex	Site	Rt./Lt.	Pattern
Urano-Suehisa et al. ⁴ (1984)	F/5	Abdomen, leg	Lt.	Linear distribution
Shirahama et al. ⁵ (1994)	M/25	Abdomen	Lt.	Linear distribution
Yokozeki et al. ⁶ (2005)	F/18	Chest, back, and arm	Lt.	Multiple linear distribution
Our case (2008)	M/27	Back, flank, and abdomen	Lt.	Linear distribution

patches appeared on his left back and spread gradually to the left flank and abdomen. After 3 months, portions of the erythematous patches became hyperpigmented. The patient complained only of intermittent itching. He was on no medications and there was no family history of hyperpigmentation. The physical examination revealed hyperpigmented patches surrounded by erythematous patches in a linear distribution along Blaschko's line (Fig. 1). Laboratory tests, including a complete blood count, blood chemistry analysis, urinalysis, and VDRL, were within the normal range or negative. Histopathologic findings showed hyperpigmentation and vacuolar degeneration along the basal layer of the epidermis. They also showed pigment incontinence and perivascular lymphocytic infiltration in the dermis (Fig. 2A). Fontana-Masson stains showed increased epidermal melanin and some pigment incontinence (Fig. 2B).

DISCUSSION

Ashy dermatosis is a chronic, idiopathic dermal melanosis characterized by hyperpigmented macules. The patches of variable shape and size are an ashen-gray color and primarily involve the face, neck, trunk, and proximal arms^{2,3}. The patient can be of any age, but the mean age of occurrence is in the 2nd decade of life^{2,7}. Ashy dermatosis is

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Fig. 1. Brown patches surrounded by erythematous patches are in a linear distribution along Blaschko's line on the left side of the trunk.

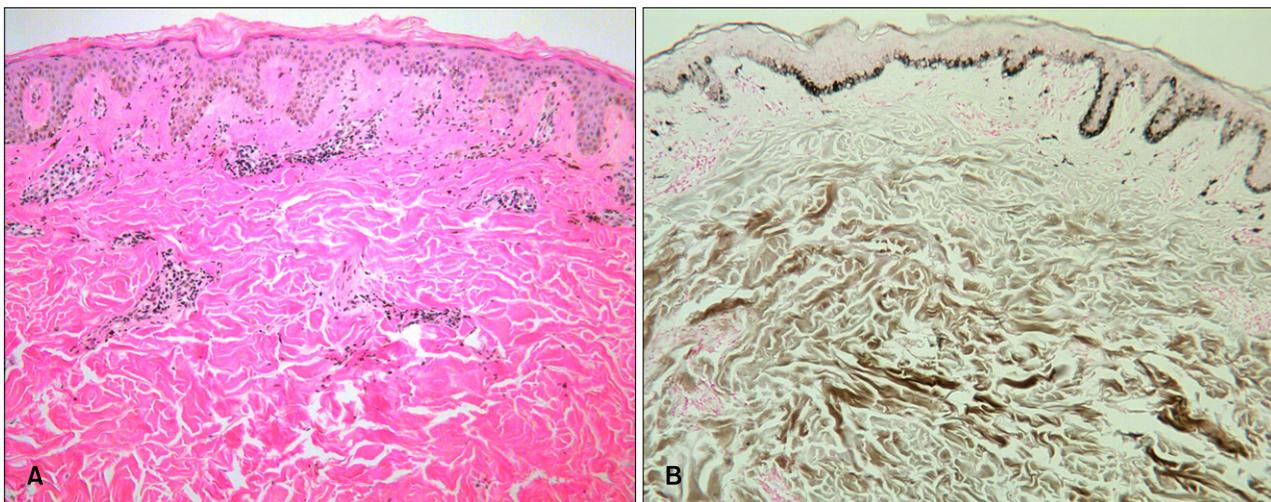


Fig. 2. (A) Histopathologic examination of the brown patch revealed mild hyperkeratosis, irregular rete ridge elongation, hyperpigmentation, basal vacuolar degeneration, pigment incontinence, and superficial infiltrate of lymphohistiocytes (H&E, $\times 100$). (B) Increased melanin granules in the epidermis and some pigment incontinence in the dermis (Fontana-Masson stain, $\times 100$).

most common in Asia and Central and South America, and is somewhat more common in women^{2,3,7}. The etiology of ashy dermatosis is unknown. Ashy dermatosis has been reported in association with the ingestion of ammonium nitrate, intestinal parasites (whipworm infection), orally administered contrast medium, cobalt allergy in plumbers, vitiligo, human immunodeficiency virus (HIV) infection, and chronic hepatitis C^{2,3,7}.

Clinically, the lesions of ashy dermatosis vary in color from slate gray to lead, and in size from 3 mm to very large confluent patches². Individual lesions can be oval, ir-

regular, or even polycyclic in shape, and continue to grow slowly. In most cases, the lesions are surrounded by an erythematous, non-scaling, slightly elevated border^{3,6}. The trunk, arms, neck, and face are commonly involved symmetrically, although there are three reports of ashy dermatosis with a unilateral distribution (Table 1)^{4,6}. Yokozeki et al.⁶ described unilateral ashy dermatosis distributed along Blaschko's lines in the first reported case in 2005. Another two cases have been reported in the Japanese literature^{4,5}. Our case is the second case of unilateral ashy dermatosis in the English literature.

The differential diagnosis of unilateral ashy dermatosis includes lichen planus pigmentosus (LPP), partial unilateral lentiginosis (PUL), and post-inflammatory hyperpigmentation (PIH)^{6,7}. LPP consists of brown-to-black patches without an erythematous border on the face and flexor folds. PUL usually presents during childhood and has no erythematous border. The histopathologic findings of prominent pigment incontinence in PIH and late stage ashy dermatosis are hard to differentiate; however, PIH can be ruled out clinically by a history of preceding cutaneous insults, such as drug or phototoxic reactions, infections, physical injury or trauma, allergic reactions, and inflammatory diseases.

Histopathologically, ashy dermatosis may show basal vacuolar changes in the epidermis, superficial perivascular lymphohistiocytic infiltration, and pigment incontinence³. A recent study reported a predominance of HLA-DR+, ICAM-1+ keratinocytes and CD8+ T lymphocytes⁸.

There is no definitive treatment for ashy dermatosis. Recently, clofazimine has been used with some success in adults, apparently because of its anti-inflammatory and immunomodulating effects⁹. A successful response to dapsone in one patient has also been reported¹⁰.

We have reported a rare case of unilateral ashy dermatosis with a linear distribution related to Blaschko's lines on the left abdomen, flank, and back. In addition, we suggest the possibility that ethnic and geographic factors are involved in the prevalence of unilateral ashy dermatosis.

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