Published online: February 25, 2011

© 2011 S. Karger AG, Basel ISSN 1662–6567 www.karger.com/cde

This is an Open Access article licensed under the terms of the Creative Commons Attribution-NonCommercial-NoDerivs 3.0 License (<u>www.karger.com/OA-license</u>), applicable to the online version of the article only. Distribution for non-commercial purposes only.

Chronic Actinic Dermatitis with Leonine Facies and latrogenic Adrenal Insufficiency Successfully Treated with Topical Tacrolimus

Ploysyne Busaracome Penpun Wattanakrai

Natta Rajatanavin

Division of Dermatology, Department of Medicine, Ramathibodi Hospital, Mahidol University, Bangkok, Thailand

Key Words

Chronic actinic dermatitis · Adrenal insufficiency · Topical tacrolimus

Abstract

Chronic actinic dermatitis is a chronic photosensitivity disorder characterized by severe eczematous lesions on sun-exposed skin areas. We report a case of chronic actinic dermatitis presenting with leonine facies and secondary adrenal insufficiency, which was successfully treated with topical tacrolimus. The facial lesions dramatically improved after sun avoidance and topical tacrolimus application. After almost 20 years of oral corticosteroid therapy complicated with secondary adrenal insufficiency, we were able to switch treatment from systemic corticosteroids to topical tacrolimus to control the patient's symptoms.

Introduction

Chronic actinic dermatitis (CAD) is a severe photosensitivity disease induced by UVB, UVA and occasionally visible light. This idiopathic photodermatosis typically affects elderly men. Clinical manifestations include eczematous lesions, infiltrated plaques and pruritic erythematous papules, which develop on light-exposed areas, particularly the face, neck and upper chest [1]. The severity of the disease in each individual depends on the degree of photosensitivity, ranging from chronic eczematous lesions to erythroderma. Histopathological findings usually show eczematous appearance but sometimes may resemble cutaneous lymphoma [2]. The diagnosis of CAD is based on the following 3 criteria: persistent photodermatitis without history of exposure to known

49

photosensitizers, abnormal delayed erythemal responses to UVB and/or UVA and/or visible light, and a histological finding consistent with photodermatitis [3]. The pathophysiology of CAD remains unclear, but current evidence suggests a delayed-type hypersensitivity reaction to endogenous photo-induced allergens. Several treatment modalities have been proposed to induce long-term remission, such as systemic corticosteroid, azathioprine, and PUVA; however, the use of these treatment options has limited efficacy and is associated with severe side effects.

Case Report

A 37-year-old male farmer presented to our hospital with a 22-year history of itchy indurated facial erythematous plaques. He has been treated with oral corticosteroids (prednisolone 30–60 mg/day) with only temporary improvement. He had no history of exposure to any chemicals, plants, or perfumes. Additionally, he had no known underlying disease and there was no family history of similar skin conditions.

Physical examination showed huge indurated lichenified erythematous infiltrative plaques on his forehead, nose, and both cheeks (fig. 1), and there were discrete erythematous papules on both forearms. A skin biopsy from a lesion of the left cheek was obtained and stained with HE. The biopsy specimen consisted of mounds of parakeratosis, spongiosis and epidermal hyperplasia associated with dense superficial inflammatory cell infiltrates, mainly lymphocytes, in the thickened papillary dermis. Routine laboratory findings were normal, except for a very low morning serum cortisol level (<1 μ g/dl). Antinuclear antibody and anti-HIV tests were negative. Patch and photopatch testing (including analysis of common photoallergens such as sunscreen, fragrance, and sesquiterpene lactone) revealed no contact sensitivity (table 1). Phototesting was done on uninvolved skin. The patient's minimal erythema dose to both UVA and UVB demonstrated a significantly reduced threshold for UVA (3 J/cm²; reference value: 30 J/cm²) and UVB-induced erythema (12 mJ/cm²; reference value: 45-60 mJ/cm²) for his skin phototype IV (fig. 2). After 4 months of topical tacrolimus therapy and sun avoidance by using sunscreen and wearing opaque clothes and a broad-brimmed hat, there was a significant improvement of his facial lesions (fig. 3). Remission of CAD could be maintained by topical tacrolimus, and the secondary adrenal insufficiency induced by exogenous corticosteroid therapy recovered after discontinuation of oral steroids.

Discussion

In this case, CAD with extensive leonine facies was successfully treated with 0.1% tacrolimus ointment twice daily. A clinically significant improvement of all lesions was observed after 4 months. There was no adverse effect except for a mild transient burning sensation during the first few treatment days. Tacrolimus is an immunosuppressive macrolide antibiotic, which inhibits calcineurin and suppresses the activation of antigenspecific T cells and the release of inflammatory cytokines, such as interleukin 2 and tumor necrosis factor [4]. T cell activation may be important for the pathogenesis of CAD. This hypothesis is supported by both the fact that the infiltrating cells are predominantly T cells and the immunological effects of tacrolimus [5]. This advocates the use of topical tacrolimus as maintenance therapy and to prevent recurrence. After a follow-up period of 1 year with strict sun protection and maintenance therapy with topical tacrolimus once daily, the patient eventually achieved remission. Although there have been several reports of CAD cases successfully treated with topical tacrolimus [5–8], this is the first case with severe and extensive leonine facies, which were uncontrolled despite 20 years of oral corticosteroid use, which in contrary induced secondary adrenal insufficiency as adverse

ARGER

50

effect. After remission, the oral corticosteroid was successfully tapered and discontinued within 6 months; thereafter, the morning serum cortisol level returned to a normal range.

In conclusion, this case demonstrates for the first time that topical tacrolimus might be an appropriate topical treatment with minimal side effects to induce and maintain remission in severe CAD cases.

Disclosure Statement

The authors declare that they have no conflict of interest.

Table 1. Photopatch test form

No. NR R NR R NR R SU1 4-tert-Butyl-4'-methoxy-dibenzoylmethane (Parsol 1789) 2.0 pet B029 □<	
SU1 4-tert-Butyl-4'-methoxy-dibenzoylmethane	
SU1 4-tert-Butyl-4'-methoxy-dibenzoylmethane Image: Constraint of the state of the stat	
SU2 4-Aminobenzoic acid (PABA) 5.0 pet A006	_
SU3 4-Isopropyl-dibenzoylmethane (Eusolex 8020)	
SU4 3-(4-Methylbenzyliden) camphor (Eusolex 6300)	_
SU5 2-Ethylhexyl-4-dimethylaminobenzoate (Eusolex 6007, Escalol 507) 2.0 pet E018 SU6 2-Hydroxy-4-methoxybenzophenone (Eusolex 4360, Escalol 567, Oxybenzone) 2.0 pet H014 SU7 2-Ethylhexyl-4-methoxycinnamate (Parsol MCX, SU8 2-Hydroxy-methoxymethylbenzophenone SU8 2-Hydroxy-methoxymethylbenzophenone <	
SU6 2-Hydroxy-4-methoxybenzophenone (Eusolex	_
4360, Escalol 567, Oxybenzone) 2.0 pet H014 SU7 2-Ethylhexyl-4-methoxycinnamate (Parsol MCX,	
SU7 2-Ethylhexyl-4-methoxycinnamate (Parsol MCX,	
SU8 2-Hydroxy-methoxymethylbenzophenone (Mexenone) 2.0 pet H020 SU9 2-Phenylbenzimidazol-5-sulfonic acid (Eusolex SU9 2-Phenylbenzimidazol-5-sulfonic acid (Eusolex SU10 2-Hydroxy-4-methoxybenzophenon-5-sulfonic SU10 2-Hydroxy-4-methoxybenzophenon-5-sulfonic Benzophenone 4) 5.0 pet H023 SU10 Thick how be title (TOC) to a tribute	—
(Mexenone) 2.0 pet H020 SU9 2-Phenylbenzimidazol-5-sulfonic acid (Eusolex 232, Novantisol) SU10 2-Hydroxy-4-methoxybenzophenon-5-sulfonic acid (Sulisobenxone, Uvinyl MS-40, Benzophenone 4) 5.0 pet H023	
232, Novantisol) SU10 2-Hydroxy-4-methoxybenzophenon-5-sulfonic acid (Sulisobenxone, Uvinyl MS-40, Benzophenone 4) 5.0 pet H023	
SU10 2-Hydroxy-4-methoxybenzophenon-5-sulfonic </td <td>_</td>	_
Benzophenone 4) 5.0 pet H023	
SUI1 Trichlorcarbanilide (TCC) 1.0 pet T013 \Box \Box \Box \Box \Box	
SU12 Tribromsalicylanilide (TBS) 1.0 pet T012	_
SU13 Musk ambrette 1.0 pet M017	_
SU14 6-Methylcoumarine (6-MC) 1.0 pet M010	
SU15 Bithionol 1.0 pet B014	
SU16 Wood mix (pine, spruce, birch, teak)	
20.0 pet Mx09	
SU17 Tetrachlorsalicylanilide (TCS) 0.1 pet T001 \Box \Box \Box \Box \Box \Box	
SU18 Hexachlorophene 1.0 pet H001	
SU19 Diphenhydramine hydrochloride 1.0 pet D021 \Box \Box \Box \Box \Box	—
SU20 Pertume mix 6.0 pet Mx08 IIIIIIIIIIIIIIIIIIIIIIIIIIIIIIIIIIII	—
SU22 Chargenthemum cinemaricafolium	—
(Pyrethrum) 1 0 pet C031	
SU23 Sesquitemente lactone mix* 0.1 net Mx18 \Box \Box \Box \Box \Box	
SU24 Lichen acid mix 0.3 pet Mx15	
SU25 Sulfanilamide 5.0 pet S010	
SU26 T026 Thiourea 0.1% pet \Box	
SU27 Ouinidire 1% pet	
SU28 Naproxen 5% pet	_
SU29 Ibuprofe 5% pet \Box \Box \Box \Box \Box	—
SU30 Diclofenac 1% pet	_
SU31 Ketoprofen 2.5% pet	
SU32 Balsam Peru 25.0 pet B001	
SU33 2-tert-Butyl-4-methoxyphenol (BHA)	_
2.0 pet B022 SU34 Chlorhexidine digluconate 0.5 aq C005	

Published online: February 25, 2011

© 2011 S. Karger AG, Basel ISSN 1662–6567 www.karger.com/cde



Fig. 1. Before treatment.



Fig. 2. Reduced threshold for UVA and UVB.



Fig. 3. After 6 months of treatment.

References

- 1 Hawk JLM, Magnus IA: Chronic actinic dermatitis an idiopathic photosensitivity syndrome including actinic reticuloid and photosensitive eczema. Br J Dermatol 1979;101(suppl 17):24.
- 2 Roelandts R: Chronic actinic dermatitis. J Am Acad Dermatol 1993;28:240-249.
- 3 Yap LM, Foley P, Crouch R, Baker C: Chronic actinic dermatitis: a retrospective analysis of 44 cases referred to an Australian photobiology clinic. Australas J Dermatol 2003;44:256–262.
- 4 Zabawski EJ, Costner M, Cohen J, Cockerell CJ: Tacrolimus: pharmacology and therapeutic uses in dermatology. Int J Dermatol 2000;39:721–727.
- 5 Uetsu N, Okamoto H, Fujii K, Doi R, Horio T: Treatment of chronic actinic dermatitis with tacrolimus ointment. J Am Acad Dermatol 2002;47:881–884.
- 6 Abe R, Shimizu T, Tsuji A, Matsumura T, Shimizu H: Severe refractory chronic actinic dermatitis successfully treated with tacrolimus ointment. Br J Dermatol 2002;147:1273–1275.
- 7 Evans AV, Palmer RA, Hawk JLM: Erythrodermic chronic actinic dermatitis responding only to topical tacrolimus. Photodermatol Photoimmunol Photomed 2004;20:59–61.
- 8 Schuster C, Zepter K, Kempf W, Dummer R: Successful treatment of recalcitrant chronic actinic dermatitis with tacrolimus. Dermatology 2004;209:325–328.