A case of recalcitrant lichen planus pigmentosus treated by oral isotretinoin

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INTRODUCTION

Lichen planus pigmentosus (LPP) is a rare dermatologic variant of lichen planus that creates substantial cosmetic distress to patients and remains a therapeutic challenge for dermatologists.[1] In cases of widespread or progressive LPP, systemic steroids, high doses of vitamin A, colchicine, dapsone, mycophenolate mofetil, hydroxychloroquine, and other systemic agents have been used but rarely result in resolution of pigmentary changes.[1-3] We report a case of a patient with recalcitrant LPP who had a remarkable response to isotretinoin therapy.

Abbreviation used:

LPP: lichen planus pigmentosus

CASE REPORT

A 46-year-old woman with medical history notable for hypertension presented to the dermatology clinic with diffuse discoloration of her face and neck. Five years prior, she noticed a new onset of dark patches on her face and neck after a few hours of unprotected sun exposure. She also felt mild burning and itching in the affected area. Medications at the time included chlorthalidone.

Physical examination found hyperpigmented, gray patches diffusely on the face, sparing the nasolabial fold and infraorbital area, extending to the neck with clear demarcation from adjacent unaffected skin areas (Fig 1, *A*). Skin biopsy of the preauricular area found subtle vacuolar interface dermatitis with marked pigment incontinence consistent with LPP and erythema dyschromicum perstans.[4]

Prior treatment trials included 9 months of hydroquinone 4% cream with topical steroids twice daily and 3 chemical peels, resulting in minimal clinical change. Chlorthalidone had also been discontinued. The patient was subsequently counseled to reduce sun exposure, use sun block, and wear protective clothing including a hat. She also went on to trial a series of various treatment regimens. Specifically, she completed a 3-month course of tacrolimus ointment 0.1% twice daily, 3-month course of hydroquinone 6%-kojic acid 6% daily, and 3-month course of hydroquinone 8%-kojic acid 6% daily, which resulted in minimal clinical response in her old lesions and the development of new pruritic lesions on the wrists, abdomen, thighs, and axilla. These new lesions on the body had ill-defined, violaceous-to-gray hyperpigmented patch morphology both with and without scale.

The patient began hydroxychloroquine, 200 mg twice daily for 6 months, with minimal improvement; she continued to have new active, pruritic lesions appearing on the trunk, wrists, and thighs with similar morphology as prior. At that time, hydroxychloroquine was discontinued, and the decision was made to trial isotretinoin, 20 mg daily, as the next course of treatment. Prior to initiation, the patient's pregnancy test was negative. Additionally, her baseline complete blood count, blood chemistry, liver function, and lipid studies were all within normal limits as were her follow-up studies. By 2 months on treatment, improvement in hyperpigmentation was observed, and stabilization of disease, with no new LPP eruptions, was achieved by 3 months. By 9 months of treatment, the patient had dramatically improved from her pretreatment appearance with near-complete resolution of her

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Fig 1. Clinical views of skin lesions before and 12 months after beginning isotretinoin treatment. **A**, Diffuse blue-gray macules and patches over the neck, forehead, nose, cheeks, and ears. **B**, After 1 year from isotretinoin treatment initiation, the lesions have dramatically improved.

bluish gray dyschromia in both her facial and body lesions equally. Her minimal residual dyschromia was brown in color. Additionally, the patient's satisfaction in her cosmetic appearance had significantly improved. At 1 year from treatment initiation (Fig 1, *B*), the patient continued to take isotretinoin, tapered down to 20 mg every other day. She had no new lesions and her residual dyschromia of prior lesions continued to fade. Low-dose isotretinoin treatment has been well tolerated with only mild, typical adverse effects of xerosis and cheilitis.

DISCUSSION

LPP commonly presents in women with insidious onset during the third to fifth decade of life and often with a prolonged clinical course. Primary morphology of LPP is blue-grey-black—pigmented macules and patches commonly distributed in photo-exposed areas. Histologic features of LPP include pigment incontinence, perivascular lymphocytic infiltrate, and basal cell vacuolar degeneration. Our patient's clinico-histopathologic features were consistent with a diagnosis of LPP.[1]

One prospective study of 32 patients with LPP by Muthu et al[5] found moderate improvement of hyperpigmentation in nearly half of patients treated with low-dose (20 mg) daily isotretinoin and sunscreen for a 6-month period. Good improvement (ie, >50%) was seen in approximately one-fifth of patients.[5] Notably, this study excluded patients with systemic therapy attempted within 3 months of enrollment and does not describe prior treatment trials of included patients. The authors also found that patients with less than 1 year of disease, those with localized disease, and those with lesions of the face and neck were associated with better clinical outcomes. Despite our patient's widespread disease and prolonged disease course of 5 years, she had near-complete resolution in her LPP dyschromia with isotretinoin treatment.

Mechanistically, isotretinoin therapy may achieve clinical efficacy in active LPP through anti-inflammatory and immune-modulating effects.[6,7] However, further investigation is needed to specifically elucidate the mechanism of action both generally in dermatologic conditions and specifically in LPP.

Our case uniquely shows an important clinical outcome from isotretinoin therapy for LPP late in its course as well as after several prior treatment failures and/or minimally responsive results. Future prospective studies are needed to investigate clinical efficacy for recalcitrant, widespread, or late-presenting LPP, as isotretinoin may be an effective treatment option for these patients.

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