Dermatology Practical & Conceptual

Graham-Little-Piccardi-Lassueur Syndrome: Favorable Response to Isotretinoin Treatment

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Introduction

Graham-Little-Piccardi-Lassueur syndrome (GLPLS) is a rare subtype of lichen planopilaris (LPP), with approximately 50 reported cases [1]. It is characterized by a triad of scarring alopecia on the scalp, non-scarring alopecia in the axillary and pubic regions, and follicular papules on the body [2-3]. Due to its rarity, treatment is based on case reports and includes topical and systemic medications, with variable efficacy. Given the lack of strong evidence, alternative therapeutic approaches are needed to prevent progression to cicatricial alopecia.

Case Report

A 33-year-old female presented with a three-year history of trichodynia, pruritus, and hair loss. Examination revealed reduced hair density at the vertex, and trichoscopy revealed loss of follicular openings, and hyperkeratosis (Figure 1). Histopathology confirmed LPP, showing perifollicular fibrosis, lymphocytic infiltration, and follicular narrowing (Figure 2). Treatment was initiated with hydroxychloroquine (200 mg every 12 hours), oral minoxidil (1 mg/day), topical clobetasol 0.05%, ketoconazole 2% shampoo, and monthly intralesional corticosteroid injections.

Despite improvement in scalp symptoms and partial hair regrowth, new follicular hyperkeratotic papules appeared on the trunk and extremities alongside patches of non-scarring alopecia in the axillary and pubic regions (Figure 1). Trichoscopy revealed yellow dots and hair thinning. These findings led to a diagnosis of GLPLS, prompting a switch from hydroxychloroquine to isotretinoin (20 mg three times weekly). After three months, the patient showed resolution of scalp symptoms, repopulation of non-scarring alopecia areas, and a decrease in hyperkeratotic papules.

Conclusions

GLPLS is the rarest form of LPP, primarily affecting Caucasian females aged 30 to 70 years [1,4]. Its etiology remains unclear, though associations with vaccines and hormonal changes have been suggested. Clinically, it manifests as a

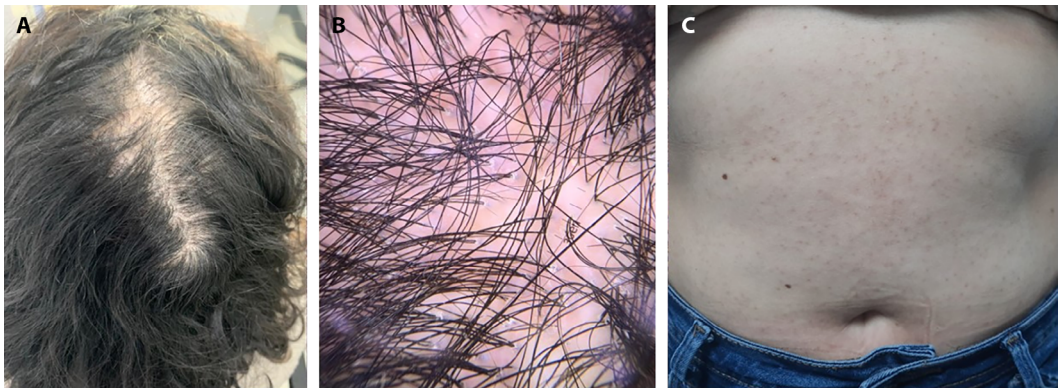

Figure 1. (A) Alopecic patch on the vertex of the scalp (B) Trichoscopy: erythematous base, loss of follicular openings, perifollicular scaling, peripilar casts, and tufted hairs (C) Hyperkeratotic follicular papules on the abdomen.

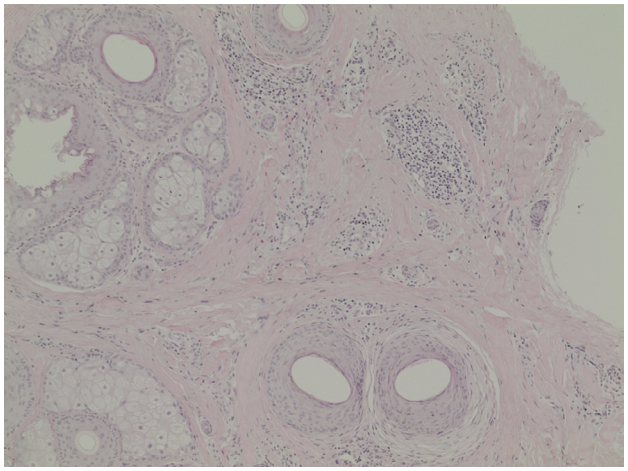

Figure 2. Hematoxylin-eosin stain 40x, decreased number of hair follicles with perifollicular fibrosis and lymphocytic infiltrate.

triad of scarring alopecia on the scalp, non-scarring alopecia in the axillary and pubic areas, and hyperkeratotic follicular papules [2,3]. Scalp findings include perifollicular erythema, scaling, and progressive multifocal alopecic patches. Trichoscopy typically reveals perifollicular erythema, fibrotic white dots, hair casts, and loss of follicular openings [5].

Histopathologically, early stages show perifollicular lymphocytic infiltration at the infundibulum and isthmus, with vacuolar degeneration and keratinocyte necrosis. Advanced stages demonstrate perifollicular fibrosis, loss of pilosebaceous units, and replacement by fibrous tracts. The disease course varies but often leads to irreversible scarring alopecia [2,3].

Treatment remains challenging and is primarily extrapolated from LPP management. The main goal is to halt disease progression and relieve symptoms. Described therapies include topical, intralesional, or systemic corticosteroids, retinoids, hydroxychloroquine, cyclosporine, methotrexate, and photochemotherapy, all with varying success rates [1,3].

Early recognition of GLPLS is crucial to initiating timely treatment and preventing irreversible alopecia. This case highlights the importance of comprehensive follow-up in LPP patients, as the GLPLS triad may develop after scarring alopecia onset. While acitretin has been used with a favorable response, to our knowledge, this is the first case where isotretinoin has been reported as a systemic therapy. Its inclusion in treatment options is valuable, particularly where acitretin availability is limited.

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