

Idiopathic Subcorneal Pustular Dermatitis in a Healthy Young Omani Female: First Report

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Abstract

Subcorneal pustular dermatosis (SPD) is a rare, chronic neutrophilic dermatosis characterized by recurrent flaccid pustules, typically affecting intertriginous and flexural areas. It is frequently associated with systemic comorbidities such as IgA monoclonal gammopathy and other immunologic or neoplastic disorders, and most often occurs in middle-aged or elderly women. We describe an unusual presentation of SPD in a healthy young Omani female with no identifiable systemic illness or immunocompromised state. The diagnosis was confirmed through clinical examination and histopathology, and the patient achieved complete remission with oral dapsone therapy. This case is notable not only for the rarity of SPD in a young, otherwise healthy individual but also for the favorable therapeutic response, reinforcing the need to consider SPD in the differential diagnosis of pustular eruptions—even in patients without underlying systemic disease. Early recognition and appropriate treatment can lead to excellent clinical outcomes.

Keywords: Subcorneal Pustular Dermatitis, SPD, pustular eruption, neutrophilic dermatosis, dapsone, Oman.

Introduction

Subcorneal pustular dermatosis (SPD), also known as Sneddon-Wilkinson disease, is an uncommon neutrophilic dermatosis first described in 1956.¹ It typically presents as recurrent sterile pustular eruptions with a predilection for flexural areas and often affects middle-aged to elderly females.^{2,3} Although its etiology remains unclear, SPD has been associated with systemic diseases such as IgA monoclonal gammopathy, multiple myeloma, and autoimmune conditions.⁴

Idiopathic cases have also been reported in the literature, highlighting its potential occurrence even in otherwise healthy individuals. Here, we present the first documented case of SPD in a healthy 30-year-old Omani woman, representing a rare and diagnostically challenging presentation and underscoring the need to consider SPD even in immunocompetent patients lacking typical risk factors.

Case Report

A 30-year-old Omani female presented with a 10-year history of recurrent pruritic pustular eruptions, predominantly involving the trunk and extremities in an annular pattern. She also reported periodic swelling of both hands and feet. The patient had no significant past medical history and was not on any long-term medications.

On dermatological examination, flaccid pustules and bullae were observed over erythematous bases, particularly over the trunk and limbs, with sparing of the face, palms, soles, and mucous membranes. Some pustules had a classical half-pustular, half-clear fluid appearance (Figure 1). There was no regional lymphadenopathy or organomegaly.



Figure 1: Numerous flaccid bullae and pustules some displaying the classical appearance of hypopyon sign; halfpustular, half-clear fluid-filled blisters situated on an erythematous base with fine scales and erosions over the trunk and the extremities.

Wound cultures from various lesions were sterile. Differential diagnoses included SPD, IgA pemphigus, pustular psoriasis, and tinea. Given the chronic relapsing course and characteristic lesion morphology, SPD was highly suspected.

Skin biopsy revealed subcorneal pustules, acanthosis, mild orthokeratosis, and dermal neutrophilic infiltrates with papillary dermal edema—findings consistent with SPD (Figure 2).

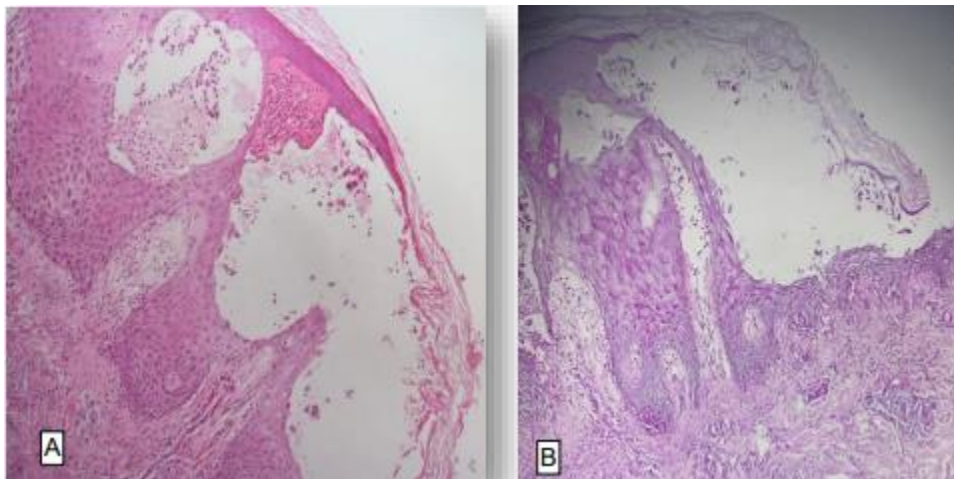


Figure 2: Acanthosis, mild orthokeratosis, the blisters are predominantly intraepidermal (subcorneal), with dermal interstitial neutrophils and papillary dermal edema.

Routine laboratory investigations, including CBC, liver and renal function tests, and bone profile, were within normal ranges. Serum protein electrophoresis was not performed; however, there were no clinical or laboratory indicators of gammopathy or systemic disease.

Direct immunofluorescence was negative for intercellular IgA, IgG, or C3 deposition, supporting the diagnosis of SPD and excluding IgA pemphigus.

Differential diagnoses were carefully excluded: IgA pemphigus was ruled out by negative DIF; pustular psoriasis was unlikely due to the absence of psoriasiform epidermal hyperplasia; and fungal infection was excluded by negative KOH and PAS stains.

The patient was initiated on oral dapsone 100 mg daily along with topical corticosteroids. At the one-month follow-up, there was complete resolution of pustular lesions, with only residual post-inflammatory hyperpigmentation remaining on both hands and feet (Figure 3). The patient expressed high satisfaction with the treatment outcome. At six months of follow-up, she remained in remission without relapse after discontinuing dapsone.



Figure 3: PIH and full resolution of active lesions noted 1 month after starting the treatment over upper and lower limbs.

Discussion

This case contributes to the expanding clinical spectrum of subcorneal pustular dermatosis and underscores its potential occurrence in young, healthy individuals without underlying systemic illnesses. SPD is classified as a neutrophilic dermatosis characterized histologically by subcorneal pustules packed with neutrophils. Although the exact etiology remains unclear, associations with systemic conditions like IgA monoclonal gammopathy and inflammatory bowel disease are well documented.^{4,6}

While SPD is typically reported in middle-aged or elderly females with systemic comorbidities, idiopathic cases in young, immunocompetent individuals have been described. These cases, including ours, emphasize the importance of considering SPD even in the absence of risk factors. The excellent therapeutic response to dapsone in this case supports its efficacy as a first-line treatment due to its anti-neutrophilic action.⁸

Unlike previously reported cases linked to immunodeficiency or hematologic malignancies,⁹⁻¹² our case illustrates that SPD can occur in otherwise immunocompetent individuals. Early diagnosis and treatment are crucial to achieving remission and preventing recurrences. The patient's sustained remission at six months reinforces the chronic but controllable nature of SPD when appropriately managed.

To the best of our knowledge, this is the first reported case of SPD from Oman, adding valuable data to the geographic and demographic distribution of the disease.

Conclusion

This case represents a rare presentation of subcorneal pustular dermatosis in a healthy young Omani woman. Early diagnosis and appropriate intervention with dapsone led to significant improvement and sustained remission. Clinicians should maintain a high index of suspicion for SPD, even in the absence of typical risk factors.

Disclosure

The author declare no conflict of interest. Written informed consent was obtained from the patient for publication.

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