Subcorneal Pustular Dermatosis Following SARS-CoV-2 Infection

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ABSTRACT
Subcorneal pustular dermatosis (SPD), also called Sneddon-Wilkinson disease, is a rare, relapsing pustular dermatosis.1 SPD has been associated with multiple myeloma, IgA Gammopathy, pyoderma gangrenosum and certain autoimmune diseases.2 However, SPD occurrence following SARS-COV-2 has not yet been reported. Herein, we report a case of SPD occurring after SARS-CoV-2 infection in a 52-year-old male. We hypothesize that the occurrence of SPD shortly following SARS-CoV-2 infection suggests the viral illness may have precipitated onset of SPD, and the patient may remain at risk for future flares of disease despite appropriate treatment and current remission status.

KEYWORDS: Subcorneal pustular dermatosis, SPD, Sneddon-Wilkinson disease

INTRODUCTION
Subcorneal pustular dermatosis (SPD), also called Sneddon-Wilkinson disease, is a rare, relapsing pustular dermatosis.1 Patients typically present with pustules on the trunk and intertriginous areas.2 It follows a relapsing-remitting course, wherein patients may experience flares for up to weeks at a time, followed by quiescence for months to years.2 Histopathology shows superficial perivascular infiltrate of neutrophils, rare eosinophils, lymphocytes, and occasional acantholytic keratinocytes.2 The etiology of SPD remains poorly elucidated, and its classification remains controversial. It has been reported in association with multiple myeloma, IgA Gammopathy, pyoderma gangrenosum and certain autoimmune connective tissue diseases.2 SPD has been reported to occur in association with several neoplastic, immunologic, and inflammatory conditions; however, it has not yet been described following SARS-COV-2 infection.3

We report a case of SPD suspected to be precipitated by SARS-CoV-2 infection, given occurrence within two weeks following infection. Since the etiology and pathogenesis of SPD remain relatively unknown, our aim is to highlight a possible connection of viral illness as one of several possible etiologies which may be associated with the development of SPD.

CASE REPORT
A 52-year-old uninsured male presented to urgent care with blistering on his torso. He was unvaccinated against SARS-CoV-2 and had tested positive approximately four weeks prior to presentation, resolution of his symptoms without intervention. The painful blisters had started approximately two weeks prior to presentation, with one to two new blisters developing each day. He had trialed over the counter hydrocortisone cream and Benadryl without significant improvement.

On physical examination, there were erythematous crusted plaques, but when looking closely, there were pustules at the periphery of the crusts. The lesions were distributed diffusely across the trunk and extensor aspects of the proximal extremities. There was no mucosal involvement, nor was there involvement of the intertriginous areas or flexural aspects of the extremities (Figure 1). Differential diagnosis included pemphigus vulgaris, subcorneal pustular dermatosis, Sweet’s syndrome, or ecthyma.

Biopsies were performed, and histopathology demonstrated neutrophils underlying the stratum corneum and within the superficial portion of the epidermis. PAS stain was negative for fungal organisms; however, yeast-like organisms consistent with Pityrosporum were identified. Within the dermis there was a superficial to mid perivascular mixed infiltrate consisting of lymphocytes, histiocytes, and neutrophils, confirming a diagnosis of subcorneal pustular dermatosis (Figure 2). On direct immunofluorescence, IgG, IgG4, IgM, IgA, C3, and fibrinogen were all negative. Bacterial culture from a mid-back pustule was negative for bacterial growth.

The patient started a 14-day course of doxycycline 100mg twice daily and a prednisone taper (60mg for 5 days, 40mg for 5 days, then 20mg for 5 days). At follow-up, the patient was no longer developing new lesions and there were scattered pink patches where the previous lesions were located. He was prescribed Dapsone and topical clobetasol 0.05% ointment; however, he stated he did not obtain these medications due to long lines at the pharmacy. In addition, he declined any further work-up as the condition had mostly resolved and because he did not have health insurance. Given the patient’s absence of insurance coverage and the substantial expenses linked to supplementary diagnostic testing, laboratory investigations primarily concentrated
on the most probable diagnosis, while alternative diagnoses were considered. On follow-up over the next year, the patient’s disease remained quiescent without treatment.

The patient has a history of basal cell carcinoma and melanoma in situ. A CT showed an incidental kidney mass, an incidental adrenal mass, and a mildly enlarged right axillary lymph node, but nothing definitive for malignancy. The masses are being followed with a series of scans but combined with a negative review of systems for other conditions including autoimmune diseases, we were not able to identify any other possible etiologies besides COVID-19.

**DISCUSSION**

Subcorneal pustular dermatosis (SPD) is a rare, relapsing pustular dermatosis with a paucity of literature regarding its underlying etiology. We present a case of SPD developing two weeks following SARS-CoV-2 infection. This is the first case report demonstrating SPD development after a proven SARS-CoV-2 infection.

Our patient presented with cutaneous lesions that developed two weeks after he tested positive for SARS-CoV-2, and were biopsy-proven to be SPD. According to the patient, he has never been diagnosed with, or experienced a flare of, SPD prior to current presentation. However, it is possible the patient may have had previous flares of mild disease and did not seek medical care.

During the coronavirus disease 19 (COVID-19) pandemic, there have been reports of exacerbation of dermatological conditions following infection with SARS-CoV-2. Flares of psoriasis and alopecia areata have been the most frequently described. Interestingly, rare cases of SPD have been described following viral infection and vaccination against viral illness. A case report from Germany in 2020 described a 51-year-old women who presented during the coronavirus disease-19 pandemic with superficial erythematous erosions surrounded by annularly arranged pustules, for which skin biopsy confirmed a diagnosis of SPD. The patient tested negative for SARS-CoV-2 by PCR. The patient’s disease was
concluded to be due to an undefined viral infection, but not SARS-CoV-2. Furthermore, a case report published in January 2023 described a 21-year-old man who developed an acute pustular and vesicular eruption consistent with SPD eight days following receipt of the Moderna COVID-19 vaccination.7,8

While the pathogenesis of SPD remains unknown, it has been classified as a form of pustular psoriasis.9 Reports describe flares of psoriasis following infection with influenza B, parainfluenza, rhinovirus, and subtypes of coronavirus.10 It has been postulated that viral infection activates toll-like receptors, including TLR3, that subsequently initiate an inflammatory cascade, resulting in overproduction of chemokines and cytokines (IL-36 and CXCL8). These processes have been linked to the psoriasis pathogenesis.10 As SPD is possibly considered a subtype of pustular psoriasis, a similar mechanism may explain our patient’s SPD flare following SARS-CoV-2 infection.

Although viral infection of the skin may play a possible role, exacerbation of dermatologic pathologies is thought to primarily be a result of systemic inflammation due to viral infection. Large quantities of cytokines are produced, which may trigger the exacerbation of immune-mediated disease. This could possibly explain the exacerbation of dermatological conditions following infection.

SPD has been considered a subtype of psoriasis and pemphigus vulgaris. A subset of patients with SPD present with cutaneous and circulating autoantibodies, particularly immunoglobulin A (IgA), against the glycoprotein desmocollin-1, found predominantly in the upper epidermis.11 Viral infections can trigger induction and exacerbation of autoimmune conditions.12 Similarly, SARS-CoV-2 virus is associated with an exaggerated immune response that could alter the vulnerable immune balance, triggering initial presentation or flare of autoimmune diseases.13 We hypothesize a similar mechanism may have resulted in onset of SPD in our patient. This data and our case support the importance of surveillance and suspicion for dermatological flares following SARS-CoV-2 infection.

References

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