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Case Report

Bruises of the mind: unmasking Gardner-Diamond syndrome in a middle-aged woman

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ABSTRACT

Psychogenic purpura, also known as Gardner-Diamond syndrome (GDS), is a rare psychocutaneous disorder characterized by the spontaneous development of painful, bruise-like skin lesions in response to emotional stress. Hereby reported is case of a middle aged-female, presenting with painful purpurae over bilateral lower limbs, with an underlying concealed history of stress. Further evaluation portrayed absence of any bleeding or clotting disorders, albeit with a positive auto erythrocyte sensitization test. Considering a diagnosis of psychogenic purpura, and after thorough psychological counselling, she depicted remarkable improvement in her skin lesions. This rare case report highlights the increased need for creating awareness amongst dermatologists about psychocutaneous diseases and emphasizes the importance of keen history taking, including an in-depth psychological evaluation. This report also underscores the role of autoerythrocyte sensitization test, making it an indispensable clinical tool in dermatological diagnosis, as every purpura does not reflect a coagulation disease or vasculitis.

Keywords: Gardner-Diamond syndrome, Purpura, Erythrocytes

INTRODUCTION

Psychogenic purpura, also known as Gardner-Diamond syndrome (GDS) or autoerythrocyte sensitization syndrome (AES) is an extremely rare condition; typically noted in women with psychiatric comorbidity.[1] Psychogenic purpura is regarded primarily as an autoimmune vasculopathy with sensitization to phosphatidylserine, a component of erythrocyte stroma.[2,3] There is role of auto-erythrocyte sensitization test (AEST) for the prompt diagnosis of this disease.[4] This condition is characterized by the sudden onset of painful purpuric skin lesions, often with a clear link to emotional or psychological distress. Severe stress and emotional trauma always precede the skin lesions.[5] We report this case of a 35-year-old female, who had a normal coagulation profile and an unremarkable systemic workup for vasculitis. A deep dive into her psychological history unveiling the underlying stress, ultimately led to the diagnosis of psychogenic purpura and she improved with thorough counselling and hand-holding.

CASE REPORT

A 35-year-old female presented with a three-year history of recurrent painful, brownish skin lesions appearing over her bilateral lower legs. She reported a spontaneous occurrence of these lesions. She denied history of spontaneous bleeding tendency, trauma, or irritant application. She was free from fever, joint pain, photosensitivity or any other systemic complaints. A scrupulous history taking revealed an underlying emotional turmoil related to her daughter's sickness, with an additional component of financial stress. She reported vague exacerbating factors pertaining to the lesions. Additionally, she denied self-infliction of lesions. The skin lesions were initially dark red, which turned brownish in color, followed by superficial peeling of skin

over a span of few days, revealing normal underlying skin. Cutaneous examination revealed multiple well defined hyperpigmented patches involving bilateral lower extremities in a symmetric distribution (Figure 2). The lesions healed with superficial skin exfoliation and underlying normal skin (Figure 3). Deep dermal tenderness was positive on few of the lesions and diascopy revealed non- blanchable purpura. Her systemic examination was unremarkable. She portrayed a persistent low mood and melancholia. Routine hematological investigations including hemoglobin, total leucocyte count, platelet counts were unremarkable. Her liver and renal function tests, as well as urinalysis were within normal limits. Coagulation profile including bleeding time, clotting time, prothrombin time, activated partial thromboplastin time, fibrinogen levels, D-dimer assay were unremarkable. Vasculitis and possibly associated connective diseases were ruled out based upon negative antinuclear antibody test and anti-neutrophilic cytoplasmic antibody test. This baffled us regarding the underlying etiology of her skin lesions. Upon re-considering the history of her underlying mental turmoil, aggravation of skin lesions during stressful situations, persistent dysthymia, and non-resemblance of the lesions to any specific dermatological cause of purpura, prompted us to consider the possible differentials of dermatitis artefacta and psychogenic purpura. To further decipher the puzzle, we performed auto erythrocyte sensitization test. We prepared a suspension of patient's own saline washed erythrocytes in a stepwise manner (Figure 1). Thereafter, intradermal injections of the patient's own washed erythrocytes, plasma and normal saline were given at 3 different sites respectively, over the left forearm. Test readings were obtained, first after 30 minutes and the next after 24 hours. The appearance and persistence of ecchymosis was obvious over the site of erythrocytes' injection, which was over and above the reading at the site of plasma- injection. The site injected with normal saline (control) did not demonstrate ecchymosis. Hence, the auto- erythrocyte sensitization test result was considered as positive in our case (Figure 4). A multidisciplinary consultation involving the psychiatry department was planned, however the patient refused, owing to the associated family stigma. Hence, after a thorough discussion with a psychotherapist, we counselled the patient regarding her dermatological condition. She was advocated ample use of topical emollients to be applied over her lesions. Detailed psychological counselling including relaxation techniques, laughter therapy, mindfulness and meditation formed the cornerstone of therapy. To our amazement, she depicted remarkable resolution of skin lesions within a week. The lesions healed with superficial scaling and underlying normal skin. The patient also underwent a consultation with occupational therapist, to cope up with financial stress management. Over the course of several months, the patient demonstrated significant improvement in her emotional well-being. She continued meditation and stress management techniques, further enhancing her ability to cope with stress and thereby preventing the recurrence of purpura.

Obtain whole blood sample in an EDTA tube
↓
Centrifuge the sample at 3000 rpm for 5 minutes
↓
Carefully discard the plasma and buffy coat
↓
Dilute the remaining RBCs in N.S. at a 1:3 ratio
↓
Centrifuge again for 5 minutes at 2000 rpm, then discard the supernatant
↓
Repeat the previous two steps twice more (a total of three cycles)

Figure 1: Steps to prepare washed RBCs is depicted in table.

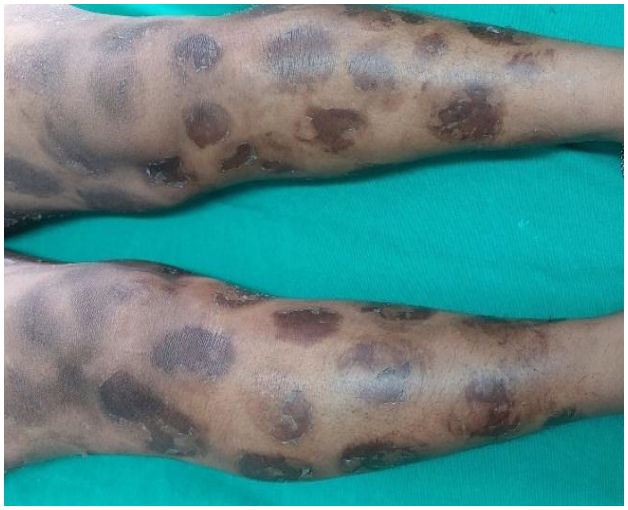

Figure 2: Multiple discrete, hyperpigmented patches involving bilateral legs in a symmetric distribution.

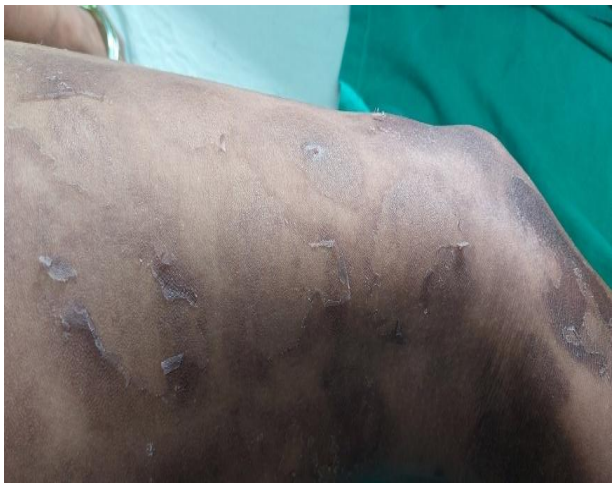

Figure 3: There is resolution of dusky lesions, with exfoliation of skin, unveiling underlying normal skin.

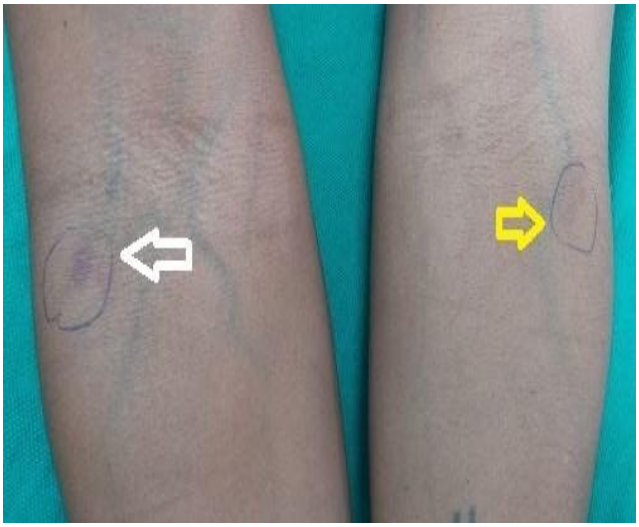

Figure 4: Right forearm with intradermal injection of erythrocyte suspension (white arrow) shows positive auto-erythrocyte sensitization test. Left forearm with intradermal normal saline injection, used as a control (yellow arrow) shows negative test.

DISCUSSION

Psychogenic purpura, also known as GDS is an infrequent condition that falls under the umbrella of psychocutaneous disorders. This entity derives its eponym from Frank Gardner and Louis Diamond, who first identified this condition. It typically presents as painful purpura, with an antecedent trigger of stress. Young females are commonly affected, although cases affecting older women and men have been described.[6] Our patient was a middle-aged female.

There are various postulated mechanisms behind pathogenesis of psychogenic purpura. The auto-immune hypothesis dwells on the formation of auto-antibodies against various components of erythrocyte stroma, including phosphatidyl serine and deoxyribonucleic acid (DNA). Psychoendocrine hypothesis revolves around neuropeptide signalling, promoting release of catecholamines and glucocorticoids.[7] Remainder of the implicated factors include stress induced oxidative damage and mast cell- degranulation, affecting red blood cell membrane, and probable role of estrogen. In our case, there was an underlying history of severe emotional and financial stress.

Clinical diagnosis of GDS is challenging, owing to the obscure nature of the disease, its pathogenesis, predisposing factors, and unreliable nature of patients' history. There is no reliable test to confirm the diagnosis and hence it is a diagnosis of exclusion, after meticulously ruling out platelet disorders, Von Willebrand's disease, hemophilia, idiopathic thrombocytopenic purpura, Henoch-Schoenlein purpura, systemic and cutaneous vasculitis.[6] The patients of GDS do not portray a history of self-infliction of lesions, contrary to dermatitis artefacta. In our case, the self-infliction of lesions was denied and coagulation disorders or vasculitis were ruled out after thorough investigations.

Auto erythrocyte sensitization test, as performed in our case, is of limited utility in the diagnosis of this condition, as positive responses to the injection of control solution and negative responses to erythrocyte injection, do not always rule out this condition.[6]

It is essential to address the psychological aspects of this disorder and employ therapies directed towards resolution of underlying emotional conflicts. Multitude of psychiatric disorders like depression, anxiety, or obsessive-compulsive disorder have been found to be associated amongst many others.[8] Our patient had dysthymia which needed prompt addressal.

Numerous medications have been proposed including vitamin C, corticosteroids, antihistamines, hormonal contraceptives and antibiotics, albeit with limited role and better responses have been observed with psychotherapy and counselling, as in our case.[6,9] Severe cases might require antidepressant medications, as against our case where we have managed the case with counselling alone.

CONCLUSION

GDS is a rare psychocutaneous disorder, manifesting as unexplained painful ecchymosis and purpura, with a typical psychological trigger. A dermatologist should harbour sound clinical acumen and a heightened vigilance for the diagnosis of this condition. Recognition of emotional stress forms the cornerstone of history taking. Albeit being a diagnosis of exclusion, prompt recognition helps avoid the exhaustive laboratory testing and unnecessary interventions. The management should be centred around thorough psychological assessment and counselling.

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