Superficial Variant of Pyoderma Gangrenosum Successfully Treated with Lower Dose Clofazimine

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Sir,

Superficial pyoderma gangrenosum (SPG) first described by Jones and Winkelmann has an indolent course, granulomatous infiltrate in histology, and no underlying systemic disease.[1,2] Here, we report a case of SPG with uncontrolled diabetes mellitus (DM) which responded well to low-dose clofazimine.

A 54-year-old female presented with painless gradually enlarging ulcers on the left breast, upper back, and left flank of one and half year duration. They failed to heal in spite of multiple courses of antibiotics. There were three non-indurated ulcers with irregular, violaceous undermined margins on the left side of the waist, breast, and upper back [Figure 1a-c]. The floor of the ulcers showed pink granulation tissue with hemorrhagic crusting. There was no lymphadenopathy.Systemic examination was unremarkable. Differential diagnoses of blastomycosis like pyoderma, atypical mycobacterial infection, deep fungal infection, and SPG were considered.

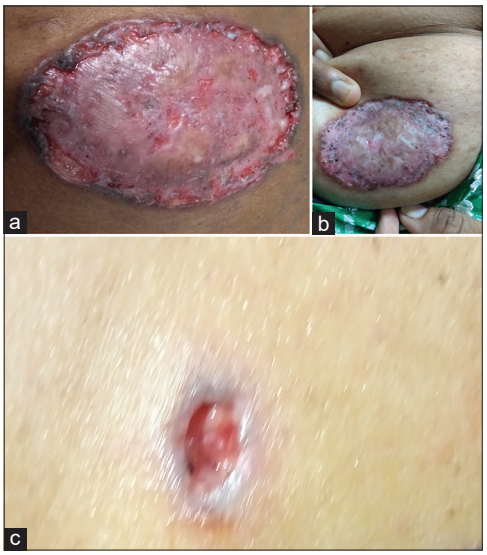

Figure 1: (a) Ulcer showing undermined edge and pale granulation tissue. (b) Ulcer on the breast showing irregular violaceous undermined margin. (c) Ulcer on the upper back showing pink granulation tissue

Pus swab yielded *Escherichia coli*, sensitive to gentamicin, cotrimoxazole, and piperacillin – tazobactum. AFB and fungal cultures were negative. Blood parameters were normal except for a raised blood sugar (FBS-230mg/dL) and elevated thyroid-stimulating hormone (11.3mIU/L). Viral markers, antinuclear antibodies, and rheumatoid factors were negative. Upper GI endoscopy and CECT abdomen did not show evidence of malignancy. Pathergy test was negative. Skin biopsy showed central necrotizing suppurative inflammation with perivascular and periappendageal lymphocytic infiltrate and leukocytoclasia[Figure 2]. Special stains for bacteria, fungus, and AFB were negative. The findings were consistent with PG though the typical granulomatous histology was lacking.

She was treated with piperacillin-tazobactum as per the culture and sensitivity report, followed by a trial of betamethasone dipropionate

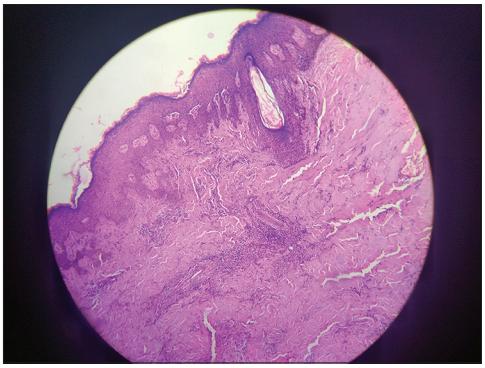

Figure 2: H and E staining(10×) showing epidermal hyperplasia, intradermal abscess perivascular, and perifollicular inflammation

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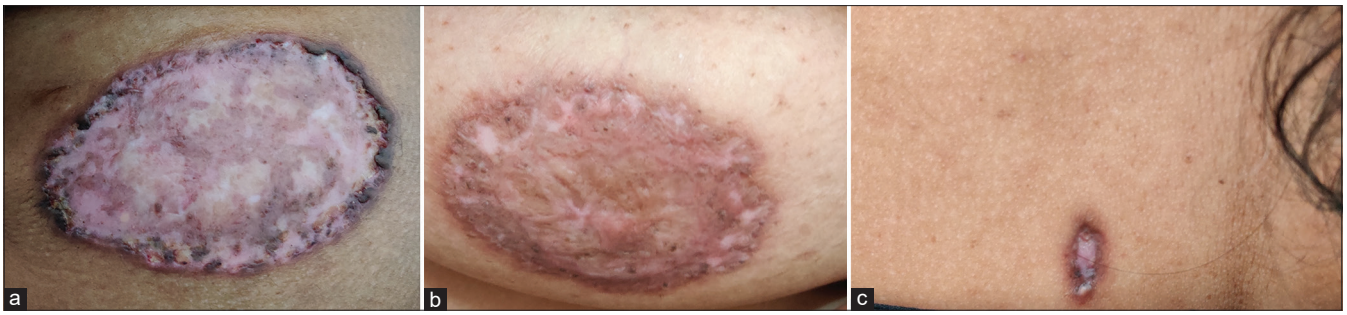

Figure 3: (a) Healing ulcer with epithelialization. (b) Healed ulcer on the breast showing cribriform scar. (c) Healing ulcer on the upper back after 4 weeks of treatment

cream**.** As the response was poor, we added clofazimine 100mg twice daily. There was a good clinical response in 10 days [Figure 3a]. The ulcer on the breast healed completely in 4 weeks, and the one on the left flank and upper back healed in 8 weeks leaving cribriform scars [Figure 3b, c]. We continued the same dose for 12 weeks and then continued at a dose of100 mg daily for another 8 weeks. No new lesions developed thereafter.

Our patient had underlying uncontrolled DM and hypothyroidism which is unusual in SPG. We prefer to term this entity as "spontaneous idiopathic neutrophilic ulcer" rather than SPG as it is clinically and histopathologicaly different from pyoderma gangrenosum (PG) (which itself is a misnomer).

Clofazimine was first successfully used in PG by Michaelsson *et al*.[3] in 1976 at a dose of 300–400 mg/day in eight cases. There are a few similar reports of SPG showing a good response to 300mg of clofazimine.Mensing*et al*.[4] reported good response at 200mg in 3 of 5 cases of PG similar to our case. However, others had used a higher daily dose.[3,5-7] We started our patient on clofazimine in view of uncontrolled diabetes, and many reports of PG showing clinical response to this agent. Although the exact mechanism of action of clofazimine is unknown, the anti-inflammatory action of clofazimine is considered pivotal for its efficacy in PG.

We emphasize the possible role of cheap and safe low-dose clofazimine in SPG in this report.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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