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Case Report

Pyoderma gangrenosum in pregnancy: a case with unusual presentation

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Abstract Pyoderma gangrenosum (PG) is an uncommon idiopathic chronic ulcerative condition, clinically characterized by initial small pustules which develop into ulcers with typical undermined borders with violaceous hue. In most cases, PG is associated with systemic diseases like inflammatory bowel disease, inflammatory polyarthritis and hematological disorders as paraproteinemia and leukemias. Diagnosis is difficult, mostly resting on the exclusion of other similar conditions, due to lack of pathognomonic histological features. We describe a case of PG in pregnancy not only for the rarity of the association, but also because of some unusual presenting features. We treated the patient by intralesional steroid injection and topical steroid application; the ulcers healed within a month.

Key words
Pyoderma gangrenosum, pregnancy, intralesional steroid.

Introduction

Pyoderma gangrenosum (PG) was first described by Brunsting *et al.*[1] in 1930. It is an uncommon chronic ulcerative condition of unknown etiology, clinically characterized by initial small pustules which develop into typical ulcers with undermined borders with violaceous hue. In most cases, PG is associated with systemic diseases like inflammatory bowel diseases, inflammatory polyarthritis and hematological disorders as paraproteinemia and leukemias.[2-3]

Diagnosis is difficult, mostly resting on the exclusion of other similar conditions, more so as the biopsy specimen does not carry pathognomonic information.[4] We describe a case of PG in pregnancy not only for the rarity of the association, but also because of some unusual presenting features.

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Case Report

A 23-year-old primigravida at 26 weeks gestation presented with a 4-week history of several painful ulcers on both groins (**Figure 1, 2**). She had been treated with topical fusidic acid cream and two courses of cefadroxil lasting a week each. Nonetheless, the ulcers continued to enlarge, the pain increased and continued to discharge purulent material. There was no history of drug intake (other than the routine iron and calcium supplements needed during pregnancy), joint pain or swelling, abdominal pain, diarrhea, constipation, weight loss. There was neither unusual fatigability nor any bruising, nor such ulceration in the past.

Examination showed several ulcers of various sizes on both the inguinal areas, ranging from 0.5 cm to 6 cm in maximum diameter. The ulcers were situated and limited to the areas of striae gravidarum which she had recently acquired. Each of the ulcers had a tender, soft, ragged and slightly raised border with violaceous hue at places.

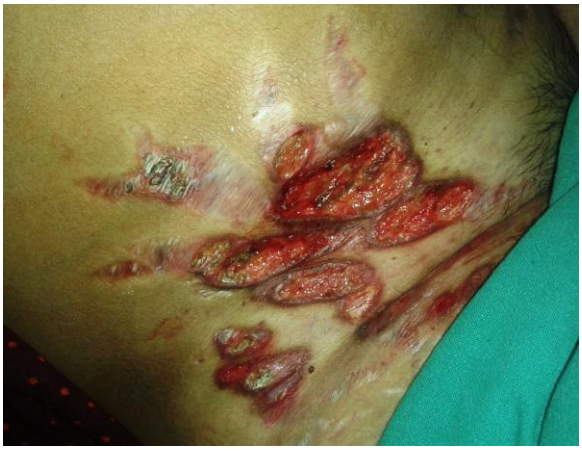

Figure 1 Several ulcers with well-defined, overhanging borders with violaceous hue, over and limited to striae distensae.

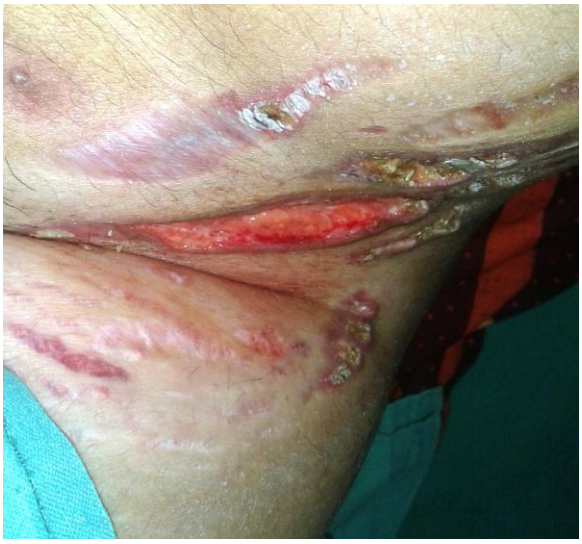

Figure 2 Same as Fig 1, left side.

Full blood count with differential was performed, which revealed anemia (10.5 mg/dl), biochemistry including hepatic and renal function were within normal limits. Serology for syphilis, hepatitis and human immunodeficiency virus (HIV) were negative. Autoantibody screen (including rheumatoid factor, antineutrophilic cytoplasmic antibodies, antiphospholipid and anti-Ro/La antibodies) was also negative. Examination of midstream urine was also non-contributory, and so were throat swabs and bacterial and fungal studies. Detailed radiological examination was not performed except for routine ultrasonography for fetoplacental profile.

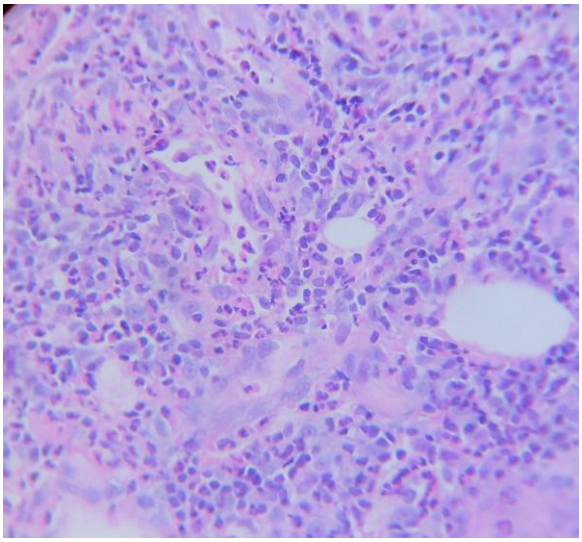

Figure 3 Neutrophilic infiltration in the dermis with abscess formation, perivascular lymphocytic infiltration along with upper dermal edema (X 400; H& E stain).

An incisional skin biopsy from the edge of the lesion was sent for histopathological examination. Examination of the slide **(Figure 3)** revealed marked neutrophilic infiltration in the dermis with abscess formation. There was perivascular lymphocytic infiltration along with upper dermal edema.

We treated the patient with weekly intralesional triamcinolone acetonide (10mg/ml) injection and application of topical mometasone furoate cream apart from regular cleaning with normal saline. The pain and ulcers reduced within a month and the patient awaits delivery.

Discussion

PG is a rare reactive condition of the skin that can cause significant morbidity and even mortality. PG is generally classified into four types: classic (ulcerative), bullous, pustular and vegetative.[2] PG is mainly a diagnosis of exclusion, using elaborate history taking, biopsy

examination and investigations including microbiological studies.

Once diagnosis is reached, treatment is generally initiated to target any underlying diseases that include inflammatory bowel disease, gammopathies, paraproteinemias, leukemias, Behcet disease, Sweet syndrome, hepatitis, HIV, systemic lupus erythematosus and vasculitides.[5]

There exists however, no current gold standard of therapy or treatment algorithm. This is again due to lack of standardization in assessment of outcome, making comparison between treatments difficult. Various therapeutic regimens have successfully been employed in PG. These include combinations of oral glucocorticoids with cyclosporine, thalidomide, methotrexate, tacrolimus, azathioprine, mycophenolate mofetil, cyclophosphamide and others.[5]

Pyoderma gangrenosum is extremely rare in pregnancy; fewer than 20 cases (pregnancy and postpartum periods included) having being reported in the international literature. Notwithstanding, there might be some possible relationship between this condition and pregnancy. The association could be explained by the pathergy phenomenon or the increase in G-CSF levels during pregnancy.[6]

The current patient was extensively investigated for associated morbidities. In the absence of the latter, we concluded pregnancy to be the etiology in this case.

Also, the localization of the ulcers on the acquired striae distensae on the abdomen suggested pathergy induced by the local stretching forces. This is another unique feature in our case, which in the best knowledge of the authors is unreported previously.

In spite of the rare occurrence in pregnancy, PG poses significant challenge to the treating physician. Since most systemic therapies are better avoided, we had to control the condition using only intralesional steroid injections and topical steroid application.

Conclusions

PG is rarely encountered during pregnancy. Most of the cases reported during pregnancy and puerperium have coexisting disease conditions, while our case had none. The ulcers in our patient were located and limited on the striae distensae on the abdomen, which is another distinctive feature.

Also, since pregnancy precludes usage of most of the available systemic therapies, our patient was managed solely on local and intralesional drug administration.

References

1. Brunsting LA, Goeckermann WH, O'Leary PA. Pyoderma (echthyma) gangrenosum clinical and experimental observations in five cases occurring in adults. *Arch Dermatol.* 1930;**22**: 655-80.
2. Powell FC, Su WP, Perry HO. Pyoderma gangrenosum: classification and management. *J Am Acad Dermatol.* 1996;**34**:395-409.
3. Prystowsky JH, Kahn SN, Lazarus GS. Present status of pyoderma gangrenosum. Review of 21 cases. *Arch Dermatol.* 1989;**125**:57-64.
4. Callen JP. Pyoderma gangrenosum: a comparison of typical and atypical forms with an emphasis on time to remission; case review of 86 patients from 2 institutions. *Medicine.* 2000;**79**:37-46.
5. Miller L, Yentzer BA, Clark A, et al. Pyoderma gangrenosum: a review and update on new therapies. *J Am Acad Dermatol*. 2010;**62**:646-54.
6. Wollina U, Haroske G. Pyoderma gangrenosum. *Curr Opin Rheumatol*. 2011;**23**:50-6.