



Pyoderma gangrenosum at an episiotomy site in successive pregnancies: A case report

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ABSTRACT

Pyoderma gangrenosum (PG) is an ulcerative, neutrophil-predominant inflammatory disease of the skin that commonly presents as painful ulcers. PG during pregnancy is extremely rare. We report the case of a 28-year-old woman with a painful, rapidly progressive ulcerative lesion at an episiotomy site presenting 16 days following a vaginal delivery. No systemic association was found after an exhaustive work-up and the patient was successfully managed with the help of oral prednisolone. It should be noted that when a postoperative ulcerative wound defect is not healing despite standard wound care, antibiotic treatment, and negative cultures, the possibility of PG should be considered. We conclude that early diagnosis of PG is essential because this condition is usually misdiagnosed and surgical intervention may lead to deterioration of lesions. The case highlights the uncommon presentation of PG at a rare site, thereby calling for a high index of suspicion in order to arrive at the diagnosis.

1. Introduction

Pyoderma gangrenosum (PG) is an ulcerative, neutrophil-predominant inflammatory disease of the skin that commonly presents as painful ulcers. PG during pregnancy is extremely rare. First described by Brunsting et al. in 1930 [1], the condition is associated with systemic diseases in at least 50% of those affected [2]. We report the case of a 28-year-old woman with a painful, rapidly progressive ulcerative lesion at an episiotomy site presenting 16 days following a vaginal delivery.

2. Case presentation

Sixteen days after a normal vaginal delivery with a right medio-lateral episiotomy, a 28-year-old woman presented with an extremely painful ulcerative lesion over the perineum. Her antenatal period had been uneventful and after spontaneous onset of labor she delivered a live girl. She developed four or five pustular lesions at the episiotomy site on the third day postpartum, which progressed to 15–20 pustules by the sixth day (Fig. 1). The lesions were associated with fever and chills. By day 10 postpartum, the lesions had progressed to form a large, painful ulcer involving rapid spread to both sides of the perineum (Fig. 2). Further enquiry revealed that the patient had developed similar lesions

at a different episiotomy site 4 days after a normal vaginal delivery two years earlier. The lesions were treated with undocumented oral medications and skin grafting. (See Fig. 1.)

On examination, the patient was pale but her vital signs were stable. On local examination, there was a single ulcer, 10 cm x15 cm, with undermined edges, violaceous rim and yellowish slough on an indurated base involving the lower part of both labia majora and fourchette extending to the perineal region. Cervix and vagina appeared healthy.

The various differential diagnoses considered were herpes zoster with secondary infection, necrotizing fasciitis, metastatic Crohn's disease and pyoderma gangrenosum. Bilateral presentation and absence of giant cells on Tzanck smear made herpes zoster unlikely. Initial investigations revealed severe anaemia with a normal peripheral smear. Ultrasound of the perineal region revealed a depth of 1.5 cm with non-involvement of fascia underneath, thereby ruling out necrotizing fasciitis. A biopsy was obtained from the edge of the ulcer and sent for histopathological examination. It revealed moderately heavy inflammation involving the whole of the dermis and also extending to the subcutaneous fat (Fig. 3).

A negative history and upper gastrointestinal endoscopy-guided duodenal biopsies showed no evidence of Crohn's disease, making that diagnosis unlikely. Rheumatoid factor and antinuclear antibody

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Fig. 2. Pustular lesions and ulceration on day 10.



Fig. 3. Partly ulcerated epidermis with moderately heavy inflammation involving the whole of the dermis and also extending to the subcutaneous fat. (H&E, 40x).



Fig. 4. Healing ulcer seen three days after the patient had started oral steroids.

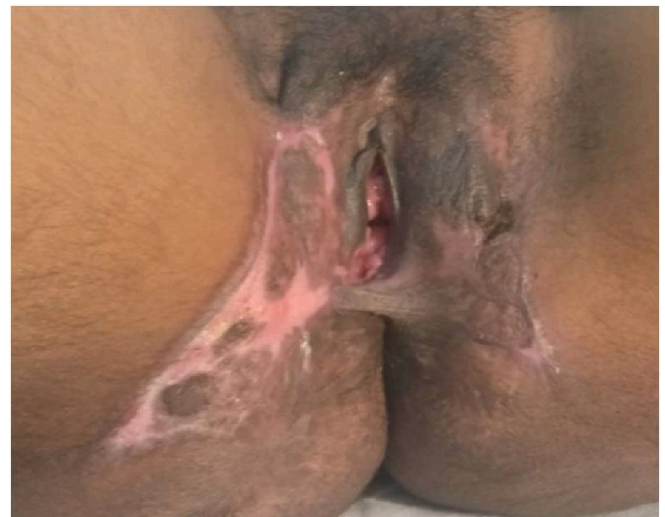


Fig. 5. Healed ulcer, three months after the patient had started treatment.

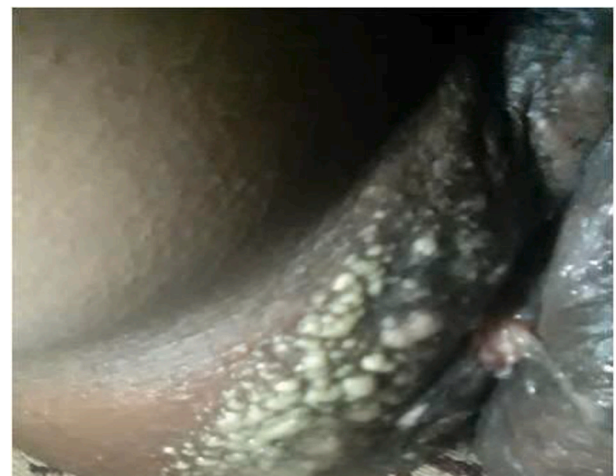


Fig. 1. Pustular lesions at the episiotomy site on day 6.

were negative. A thyroid function test and ultrasound of the whole abdomen revealed no abnormality.

The patient was started on oral prednisolone at 1 mg/kg/day. There was remarkable improvement within 2–3 days and the ulcer started healing (Fig. 4). No debridement was done to avoid pathergy reaction. Local compression dressing with hydrogen peroxide was done twice daily.

The patient received adequate analgesia and remained afebrile throughout her stay. On follow-up, the lesions had healed completely and oral steroid was tapered over a period of 3 months (Fig. 5).

3. Discussion

Pyoderma gangrenosum is a rare, neutrophilic dermatosis. Most patients are aged 20 to 50 years, and women are more often affected than men. Lesions can develop spontaneously, after surgery or after minor trauma [3]. The latter form is known as the pathergy phenomenon, and is seen in up to a third of cases [4]. In 50% of patients, comorbidities such as inflammatory bowel disease, haematological disorders and arthritis are present [5]. With this in mind, upper gastrointestinal endoscopy was employed in the present case. The incidence of pyoderma gangrenosum has been reported to be about 3–10 cases per million people per year [6], making it highly rare in

pregnancy.

The pathergy phenomenon or altered neutrophil function has been found to be an important etiological factor for PG in pregnancy, without any underlying systemic disease. This is explained as potential etiology of the neutrophil-predominant, inflammatory response of PG occurring after individuals have experienced trauma to skin during pregnancy. Biopsy samples taken from an ulcer edge show a deep suppurative folliculitis with dense neutrophilic infiltrate on histopathology [7].

Treatment options include topical therapies like local wound care and dressings, topical cromolyn sodium 2% solution, and 5-aminosalicylic acid [8]. The treatment of choice is systemic high-dose corticosteroids. Immunosuppressive therapy with cyclosporine, dapsone, infliximab, intralesional cyclosporine, azathioprine, chlorambucil, mycophenolate and several other drugs have been used successfully [9]. When there is extensive tissue loss, autologous split skin graft can be tried, but the disease can flare up at the donor site. Use of bioengineered skin, like the dermal regeneration template Integra, hair follicle stem cell-derived autologous keratinocyte sheets Epidex, or hyaluronic acid-based autologous keratinocyte delivery system laser-skin, is still experimental [10].

We conclude that early diagnosis of PG is essential because this condition is usually misdiagnosed and surgical intervention may lead to deterioration of lesions. When a postoperative ulcerative wound defect is not healing despite standard wound care, antibiotic treatment, and negative cultures, the possibility of pyoderma gangrenosum should be considered.

Contributors

Snigdha Rao contributed to the conception and design of the study, and interpretation of data, and was directly involved in the care of the patient.

Vanita Jain was involved in revising the draft report critically for important intellectual content, and was involved in the direct care of the patient. Rajsmi Bhattacharjee contributed to the drafting of the article and revising it critically for important intellectual content.

Vikarn Vishwajeet contributed to the drafting the article and revising it critically for important intellectual content. Geetika Thakur contributed to the conception and design of the study, and interpretation of data.

Conflict of interest

The authors declare that they have no conflict of interest regarding the publication of this case report.

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Patient consent

Informed consent was taken from the patient before sending the case report for submission.

Provenance and peer review

This article was peer reviewed.

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