Case Reports in Women's Health 22 (2019) e00102

Contents lists available at ScienceDirect

Case Reports in Women's Health

journal homepage: www.elsevier.com/locate/crwh

Perineal pyoderma gangrenosum in pregnancy: A case report

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ARTICLE INFO

Article history:
Received 15 January 2019
Received in revised form 12 February 2019
Accepted 14 February 2019

Keywords:
Pyoderma gangrenous
Pregnancy
Perineal region

ABSTRACT

Pyoderma gangrenosum is a rare ulcerating neutrophilic dermatosis. We describe the case of a 28-year-old woman with pyoderma gangrenosum in the perineal region during pregnancy. Cytological analysis of a skin biopsy specimen showed neutrophilic infiltrates across all the layers of the dermis, confirming the diagnosis of pyoderma gangrenosum. Determining a management plan, including the mode of delivery, was difficult. Oral prednisolone was started and her ulcer started to improve, but she still had the ulcer when she reached full term. Because there was a concern that the ulcer would be worsened by vaginal delivery, cesarean section was performed. After her delivery, pyoderma gangrenosum had not appeared at the cesarean incision and the ulcer in the perineal region had improved. Obstetricians should be aware of pyoderma gangrenosum as a differential diagnosis when vulvar ulceration develops during pregnancy.

1. Introduction

Pyoderma gangrenosum (PG) is a rare ulcerating neutrophilic dermatosis. It is typically affects the lower limbs and occurs after minor trauma or a surgical procedure. PG during pregnancy has been previously reported [1], and in cases have been reported of PG developing in the region of a cesarean scar [1–4]. We describe a case of perineal PG region during pregnancy. Obstetricians had difficulty in diagnosing PG and determining a management plan, including the mode of delivery.

2. Case report

A 28-year-old nulliparous woman presented at 33 weeks of gestation complaining of painful ulcers in the perineal region. She had experienced these symptoms for two weeks. At clinical examination an ulcer 2 cm in diameter was noted in the right perineal region. The ulcer was associated with redness and edema in the surrounding region and white fur at the ulcer mound. At the time of examination, bacterial ulceration of the vulva was suspected and empirical treatment with gentamicin ointment was started. After 1 week (at 34 weeks, 4 days of gestation), the ulcer had increased in size and was causing intense pain. Tissue cultures for bacteria and fungi were negative.

Genital herpes can cause pain, itching and sores in the genital area. Because of suspected herpes infection, treatment with vidarabine ointment was initiated, but examination of a skin scraping found no infection with herpes virus. Hematological analysis was negative for immunoglobulin M and positive for immunoglobulin G of herpes simplex virus, which indicated that active infection with herpes virus was not present. One week later (at 35 weeks, 5 days of gestation), the ulcer had increased to 5 cm in diameter and had infiltrated more deeply. Furthermore, similar lesions were noted in her left labium minus, which further increased the pain (Fig. 1).

Dermatological assessment was instigated and PG on the basis of the characteristics of the ulcers was suspected. A biopsy of the wound margin showed neutrophilic infiltration across all the layers of the dermis, which confirmed the diagnosis of PG. Oral administration of prednisolone 15 mg daily was started at 36 weeks and 4 days of gestation. At that time, it was discovered that the patient had a vulvar ulceration several years previously that had resolved with topical application of prednisolone. Again the patient responded to prednisolone and at 37 weeks and 6 days of gestation the ulcer floor had flattened (Fig. 2). Because of concern that this ulcer in her perineal region might worsen during vaginal delivery, cesarean section was recommended. Elective cesarean section was conducted at 38 weeks 0 days of gestation and a healthy female neonate was delivered.

The surgical procedure was uneventful. Five days after the operation the ulcer had an even shallower and flatter mound. The pain had also improved, leading to a reduction in the dose of prednisolone to 5 mg daily. Because the course was favorable, she was discharged with her

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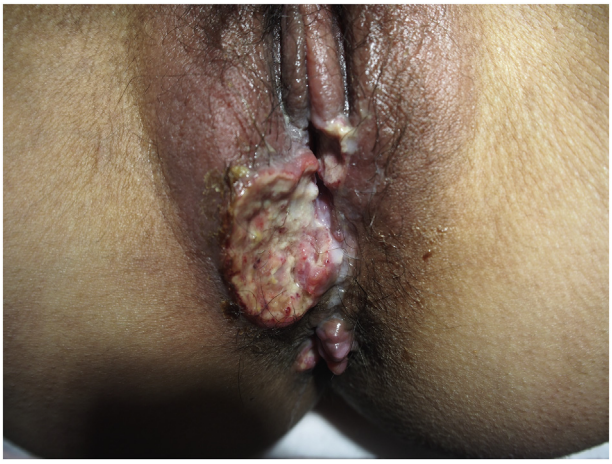

Fig. 1. The wound in the perineal region before treatment for pyoderma gangrenosum. Appearance of the ulcer was associated with redness and edema in the perineal region.

neonate without any complications six days after surgery. On postpartum day 30, the ulcer had further regressed (Fig. 3). No abnormal findings were observed at the site of the cesarean section. Prednisolone was discontinued and the ulcer had not recurred at two-month follow-up.

3. Discussion

PG is one of the neutrophilic dermatoses. It often begins as pustules, erythema, and blisters, which then rapidly expand in a centrifugal pattern to form a well defined ulcer. The ulcer is characterized by a raised border with associated erythema and edema in the surrounding region [5]. PG typically affects the extremities [6] and occurs after minor trauma or a surgical procedure, sometimes even spontaneously.

The prevalence of PG is approximately 3–10 patients per million population and occurs mainly between the ages of 20 and 50 years [7]. Women are more frequently affected than men [7]. PG during pregnancy is extremely rare.

PG often occurs in association with a systemic condition such as inflammatory bowel disease, rheumatological disease, paraproteinemia, or hematological malignancy [5]. To what extent pregnancy may increase the risk of PG is unclear. However, abnormalities of the immune system during pregnancy may affect the immunological response, possibility triggering PG [8].

Because PG does not show any specific clinical laboratory or pathological findings, it is diagnosed on the basis of clinical symptoms and course. Biopsy is useful to exclude other pathology, such as malignancy and infection. Although half of patients with PG present with only dermatological symptoms, these patients should be examined for complications, depending on their medical condition.

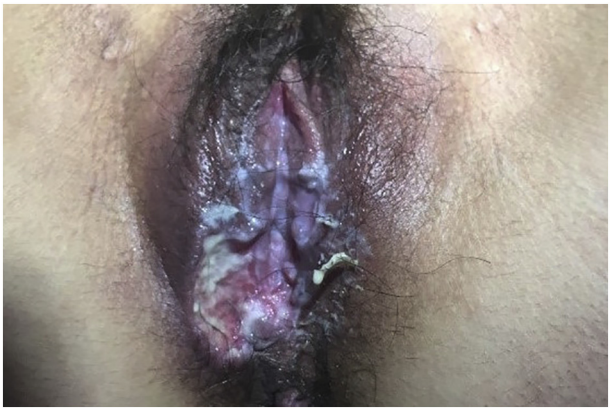

Fig. 2. Appearance of the wound on the 15th day of hospitalization. Pyoderma gangrenosum in the perineal region responded to prednisolone with a flattened ulcer floor.

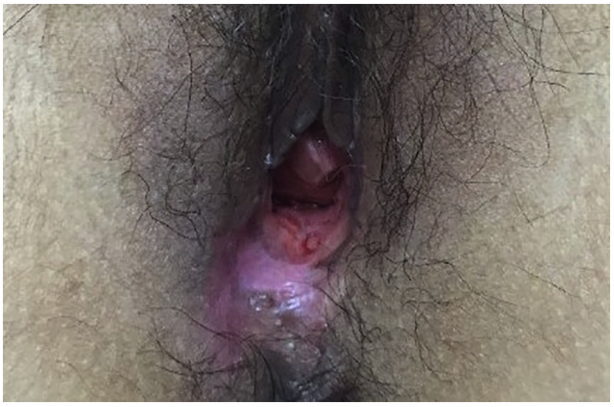

Fig. 3. By postpartum day 30, the ulcer had further regressed, with a shallower and flatter ulcer mound after treatment.

Several case reports of PG during pregnancy have been published, but many of these patients developed the disease in the lower legs or chest [1], [2], [9] [10]. Additionally, there have been multiple reports of PG occurring at the site of a cesarean incision [3], [4], [8], [10] and at the site of episiotomy during the puerperal period [11]. With regard to PG of the perineal region, a previous case report described PG at episiotomy sites [11]. However, there have been few case reports of PG in the perineal region during pregnancy, as in the current case.

The approach to treating PG during pregnancy is similar to treatment of PG in patients who are not pregnant. However, currently there is no standard treatment for PG [12]. Treatment is with immunosuppressive agents, such as high-dose corticosteroids or cyclosporine A. For pregnant women with PG, there is a need to control disease progression during delivery and postpartum.

Park et al. concluded that the best way to prevent PG is to avoid traumatic lesions [4]. In this respect, vaginal delivery might be considered better than cesarean section. Cesarean delivery was chosen in the current case because the patient still had a deep ulcer in the perineal region at 37 weeks of gestation. If a cesarean incision is made when no diagnosis has been made and no treatment has commenced, PG could develop at the wound site. Therefore, early diagnosis and treatment are especially desirable with pregnant patients. In our case, acute treatment was delayed because the cause of the ulcer was initially presumed to be infection with bacteria or herpes virus. If a patient presents with an ulcer in the perineal region during pregnancy, prompt consultation with a dermatologist for consideration of the possibility of PG is recommended, even though such cases are rare.

Contributors

All authors made a substantive contribution to the information and material submitted for publication.

Conflict of interest

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

Funding

No funding was sought or secured in relation to this case report.

Patient consent

Informed consent was obtained from the patient for the publication of this case report and the accompanying images.

Provenance and peer review

This case report was peer reviewed.

Acknowledgements

We thank Ellen Knapp, PhD, from Edanz Group (www.edanzediting.com/ac), for editing a draft of this manuscript.

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