CASE REPORT

Pyoderma gangrenosum induced by episiotomy

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SUMMARY

We present a patient who developed genital ulceration within hours following episiotomy procedure during a normal vaginal delivery. This was initially treated by the gynaecology and medical team as cellulitis with no improvement. A diagnosis of pyoderma gangrenosum (PG) was made by the dermatology team 12 days later. On further investigation, she was found to be hepatitis C positive. We report this case to highlight the phenomenon of pathergy and frequent misdiagnosis of PG by other medical teams. When a postsurgical wound is not healing despite relevant systemic treatment, the clinician should suspect PG as an early diagnosis and treatment is crucial.

BACKGROUND

Pyoderma gangrenosum (PG) is a chronic ulcerative cutaneous dermatosis with distinctive morphological features and often clinically diagnosed. To the best of our knowledge, there are no reported cases of genital pyoderma gangrenosum (PG) arising at the site of episiotomy in postpartum period in a patient with hepatitis C virus (HCV) infection.

CASE PRESENTATION

A 29-year-old Romanian woman, primigravida with an uncomplicated pregnancy, had a spontaneous vaginal delivery. Two hours after an episiotomy procedure, she developed a painful rash along the perineal area at the surgical site. It progressed rapidly with subsequent painful erosions and ulcerations. She was managed by the obstetrics and gynaecology team for a presumed post episiotomy bacterial infection with intravenous piperillin/tazobactam, clindamycin and metronidazole. Later, on the advice of the microbiology and infectious disease team, acyclovir was added to cover for possible herpetic infections. Skin swabs for viral and bacterial cultures were negative. Her skin continued to deteriorate, associated with severe pain requiring regular paracetamol and morphine-based analgesia. Since there was no improvement, at day 12 postpartum, dermatology input was sought.

Clinical examination revealed tender erosions and ulcerations with marked surrounding erythema and violaceous edges involving the perineum extending to the natal cleft abutting the episiotomy site (figure 1). A clinical diagnosis of PG was made and further investigations were requested.

INVESTIGATIONS

There are many recognised associations with PG, including rheumatoid disease, inflammatory bowel disease and myeloproliferative diseases

such as acute myeloid leukaemia. These warrant investigation and may also influence the choice of treatment.

Laboratory investigations showed elevated hepatic transaminases; aspartate aminotransferase and γ -glutamyl transferase at 470 iu/L (N: 10–50). Antibodies to hepatitis B surface and core antigens were negative, but hepatitis C antigen was positive with a mild to moderate viral load, which was a new diagnosis. Serological tests including HIV, syphilis, toxoplasmosis, herpes simplex, cytomegalovirus and rubella were negative. Other tests, such as vasculitic screen, rheumatoid factor, anti-nuclear antibodies, antineutrophilic cytoplasmic antibodies and extractable nuclear antibodies, cryoglobulins, serum protein electrophoresis, complement levels and Quantiferon test, were all normal. Skin swabs for bacterial and viral cultures were negative.

Skin biopsy from the edge of an ulcer showed focal ulceration of the epidermis associated with fibrin purulent surface exudate. Superficial dermis is markedly oedematous. On deeper levels, moderate numbers of neutrophils were found in the dermis. There was no tissue necrosis or vasculitis. Stains for bacterial and fungal organisms were negative. Histological features were consistent with PG.

Liver ultrasound showed multiple calculi in the gallbladder with some fatty liver infiltration. *Helicobacter pylori* was positive with evidence of chronic gastritis on gastroscopy. While colonoscopy did not show any evidence of inflammatory bowel disease.

DIFFERENTIAL DIAGNOSIS

- Post episiotomy cutaneous bacterial infection
- Pathergic response PG
- Erosive lichen planus
- Herpes simplex infection
- Cutaneous vasculitis

TREATMENT

Efficacy of systemic oral corticosteroids is well recognised treatment for PG, but in milder cases topical potent steroids may also be beneficial. Our patient was started on prednisolone (0.5 mgs/kg), oral omeprazole 40 mgs daily and clobetasol ointment with standard wound care. Her symptoms of pain improved dramatically over the next 48 h. At day 7, she developed severe epigastric pain and subsequently corticosteroids were stopped. An oesophago-gastroduodenoscopy showed that the gastritis was most likely induced by oral steroids, which were treated with intravenous omeprazole. Her skin started to flare with new areas of erosions in the

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Figure 1 Displays well-demarcated violaceous erosions and ulceration secondary to pyoderma gangrenosum postepisiotomy. The importance of this image is that pyoderma gangrenosum is a clinically diagnosed entity presenting as pustules which enlarge forming ulceration with an undermined margin. Any skin site can be affected.

perianal area over the next 3 weeks. Restarting oral corticosteroids and adding immunosuppressant agents at this stage was challenging in view of her current gastric symptoms and HCV.

Her case was discussed at a multidisciplinary team meeting and a treatment with cyclosporine or biologics was deferred. She was started on a lower dose of prednisolone (0.3 mg/kg) and mycophenolate mofetil 750 mg two times a day (later increased to 1000 mg two times a day). After 8 weeks, there was no significant improvement. The disease was affecting her daily life activities and she was finding it extremely difficult to cope and caring for her newborn baby. Dapsone 50 mg two times a day was added, and for the first time in 3 months, there was a significant improvement.

OUTCOME AND FOLLOW-UP

PG healed completely over the next 4 weeks (figure 2). At present, she is due to be reviewed by the Hepatology department with a view to starting hepatitis C therapy.

DISCUSSION

PG is a rare ulcerative non-infectious inflammatory neutrophilic dermatosis, which can be extremely painful.¹ As with few other skin diseases, PG is likely to be encountered by other hospital specialties and early dermatology input should be sought. Delayed or mis-diagnosis can lead to progression of the disease causing detrimental effects on the patient's morbidity. As in our case, misdiagnosis of PG as a bacterial infection is reported in the literature.² Skin biopsies are not specific as they rarely yield characteristic features but are helpful in excluding other causes of cutaneous ulcerations.³

Figure 2 Complete healing of pyoderma gangrenosum.

Up to 30–40% of patients' PG can be initiated and aggravated by minor trauma, debridement or surgery, a phenomenon known as pathergy.^{3–4} Postoperative PG has been reported most commonly after abdominal surgery, breast surgery or venepuncture, but there is no reported case of PG after episiotomy.⁵ When resulting from operative wounds, PG usually begins as multiple small areas of dehiscence or ulceration several days to a few weeks later.

Common associations with PG are inflammatory bowel disease, arthritis and haematological malignancies.³ Rarely reported associations are with liver diseases including chronic active hepatitis, primary biliary cirrhosis and sclerosing cholangitis.⁶ Association of PG with HCV is rare but was reported previously.^{6–7} In this series, PG associated with HCV infection treated with pegylated interferon α -2a and ribavirin had a varied outcome of PG with one case resolving while the other flared.¹ All patients with suspected PG should have a full workup, including a thorough history, to rule out other differential diagnosis and look for possible associations, as in our case, revealing a chronic HCV infection. Unusual presentations of PG involving the genital region have been described before, but in a recent retrospective review of 103 patients with PG, lower leg extremities were the most common site while there was no genital PG recorded in the cohort.¹

Our case had a significant physical and psychological burden affecting all facets of her life including social activities, emotional well-being and, to a greater extent, transition into motherhood. In a retrospective review by Binus *et al*,¹ depression was noted to be prevalent. In addition to treating the underlying associated disease, the mainstay of treating PG requires immunosuppression therapy. There is no single effective treatment therapy for PG, but oral steroids have proved to control the disease quickly and could be continued until PG is healed.^{1–3} In our case, PG was refractory and required immunosuppression with steroid sparing agents. The selection of immunosuppression therapy was challenging in view of HCV status. Dapsone and minocycline are frequently prescribed agents, while mycophenolate mofetil has been reported to have a good safety profile and is effective in treating PG.³ Newer immunomodulating therapies are increasingly being used in treating PG; these include infliximab, etanercept and adalimumab, but further studies are required to assess their efficacy.⁴ To the best of our knowledge, there are no reported cases of genital PG arising at the site of episiotomy in a patient with hepatitis C in the literature.

Learning points

- ▶ If a postsurgical wound is not healing, consider pyoderma gangrenosum (PG) as a diagnosis and actively seek dermatology opinion.
- ▶ PG can be associated with inflammatory bowel disease, arthritis and haematological malignancies.
- ▶ Rarely reported associations are with liver diseases including chronic active hepatitis such as hepatitis C, primary biliary cirrhosis and sclerosing cholangitis.

Contributors AA and MS made substantial contribution to the conception and write-up of the case report. BR drafted and edited the article. KA drafted the article and made revisions.

Competing interests None declared.

Patient consent Obtained.

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