

Post-Partum Pyoderma Gangrenosum Following a Cesarean Section

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Abstract

The occurrence of Pyoderma gangrenosum (PG) during pregnancy or postpartum has rarely been reported. These forms can also occur on a recent scar facilitated by the pathergy phenomenon.

A 25-year-old woman showed an abruptly worsening of a post-caesarean wound that evolved rapidly into a painful, purulent, extensive ulcer with breakdown of sutures, along with fever and leukocytosis despite antibiotic therapy.

The diagnosis of postoperative PG was evoked on the unusual course of the surgical wound, the existence of old cribriform scars on the legs, the clinical appearance of the ulceration around the surgical incision and confirmed by histology, as well as the favourable outcome under systemic corticosteroid.

This case raises once again the issue of the possible relationship between pregnancy and PG. In our case, pregnancy could have triggered the PG, and the Caesarean section facilitated its clinical expression by the pathergy phenomenon. Thus, pregnancy could be in the same way as chronic inflammatory diseases, such as arthritis, inflammatory bowel disease and haematological disorders, a triggering or exacerbating factor of PG.

Keywords: Pyoderma gangrenosum; Pregnancy; Pathergy

Introduction

Pyoderma gangrenosum (PG) is a rare disease, belonging to the spectrum of neutrophilic dermatoses [1]. It can be associated in up to 50% of cases with an underlying systemic condition, such as inflammatory bowel disease (IBD), rheumatologic disorders and haematological malignancies [1]. The occurrence of PG during pregnancy or postpartum has rarely been reported [2,3]. These forms may also occur on a recent scar facilitated by the pathergy phenomenon [2]. We report a case of patient with a recurrence of a PG during pregnancy on a Caesarean incision (CS).

Case Presentation

A 25-year-old woman had a Pfannenstiel caesarean sectional incision following an umbilical cord prolapse that occurred during the delivery of her first pregnancy at Aristide LeDantec University Hospital.

At the 4th day, after CS, the woman complained of persistent and severe pain on the surgical site with purulent discharge and a high-grade fever (38.2°C). Despite a negative bacteriological skin swab, the patient was treated with parenteral antibiotic therapy (Amoxicillin-clavulanic acid and Metronidazole) in view of a likely bacterial superimposed infection of the surgical wound.

After 3 days of treatment, she presented an alarming worsening of the wound with rapid increase in its size and wound dehiscence of the sutures at the CS site. There was a rapidly forming, large painful, necrotic ulceration with a purulent floor.

In view of this very unusual development of the wound and the presence of old scars on her legs, a dermatological consult was requested.

The physical examination revealed a good general condition and a temperature of 38.1°C.

There was a large and deep ulceration, around the CS wound, with characteristic violaceous undermined edges. (Figure 1). There were cribriform scars in the lower limbs (Figure 2). These scars are secondary to a PG treated 4 years ago with prednisone.

Laboratory investigations demonstrated a high white blood cell count (WBC) of 12,110/mm³ with neutrophilia (8400/mm³). Inflammatory



Figure 1: Wound dehiscence of the sutures at the caesarean incision site, leading to a large, deep and extensive ulceration.



Figure 2: Cribriform scars in the lower limbs.

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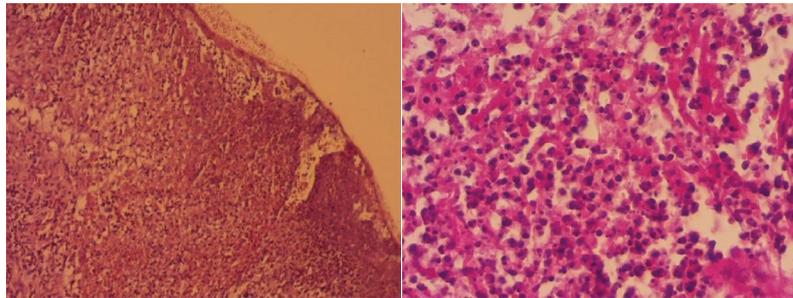


Figure 3: Dense dermal infiltrate of neutrophils with epidermal necrosis (Hematoxylin Eosin 40x).

syndrome with serum C-reactive protein at 48 mg /L and erythrocyte sedimentation rate (ESR) at 90 mm/hour. The bacterial culture revealed negative result. The skin biopsy revealed a dense dermal infiltrate of neutrophils with epidermal necrosis (Figure 3). The diagnosis of post-surgical PG was subsequently made. No associated disease with the PG was found after digestive and rheumatological explorations. The patient was treated with oral prednisone (1 mg/kg/day) and standard wound care. The clinical course was characterized by a stable apyrexia from the fourth day of treatment with a disappearance of the inflammatory syndrome. Complete healing was achieved after 32 days of treatment.

Discussion

PG is a rare neutrophilic dermatosis whose frequency is estimated to be between 3 and 10 cases per million inhabitants per year [4]. The condition usually affects adults, with no predilection for sex, race or geography [5].

Clinically, the PG is usually characterized by one or multiple painful ulcers with centrifugal extension, without satellite lymphadenopathy, localized at various sites of the tegument and sometimes accompanied by fever and leukocytosis.

The pathogenesis of PG is unknown however, a dysfunction of neutrophilic chemotaxis under the influence of cytokines particularly interleukin 8 has been incriminated [6]. PG is also characterized by a pathergy phenomenon which is found in 50% of cases [1]. Pathergy is an exaggerated skin reaction to minor trauma. Su et al. believe that this is an inadequate cellular response to an antigenic modification of the skin by trauma [1]. This phenomenon explains the preferential location of the PG for the lower limbs. This pathergy phenomenon can sometimes be linked with a surgical incision [7].

In spite of this, post-surgical PG (PSPG) is rare. In a review of the literature over the past 7 decades (January 1946 - June 2013), only 220 cases have been reported [8]. In this review, PG resulted from breast surgery in 25%, cardiothoracic and abdominal in 14% respectively and obstetric in 13% of the cases [8].

PSPG results in aggravation of the operative wound which becomes hollow, painful and purulent, simulating from all points of view an infectious cause. However, it should be mentioned that antibiotics are ineffective and surgical debridement worsens the condition due to a pathergic response [7-9]. In our case, the diagnosis of PSPG was made based on the unusual flare-up of the operative wound, the existence of old cribriform scars on the legs suggestive of a previous PG and the clinical appearance of the ulceration around the surgical incision. Subsequently, it was confirmed by histology, and further supported by the favourable resolution with the use of systemic corticosteroids. Indeed, the presence of cribriform scarring constitutes a high index of suspicion for the retrospective diagnosis of PG.

PG can be isolated or associated in 50% of cases with underlying inflammatory conditions, such as inflammatory bowel disease (IBD), rheumatoid arthritis and hematologic disorders [10]. In our patient, no associated disease was found. In the medical literature, cases of PG related to pregnancy have been reported [2,3]. These were PG that occurred or recurred during pregnancy or postpartum [3,11]. Most of these cases were not associated with underlying pathology, and pregnancy appeared to be the only triggering factor [3,12]. Of these cases, the PG sometimes appeared on a recent scar [13]. Our observation, as well as other cases of pregnancy-induced PG described in the literature, suggest that there is a link between pregnancy and PG. Pregnancy may be a triggering or promoting factor in the development of PG. In our observation, it seems that the pregnancy has triggered the recurrence of the dermatosis, and that the caesarean facilitated the clinical expression by the pathergy phenomenon. Indeed, neutrophil activation could be promoted by the significant increase in granulocyte colony stimulating factor (G-CSF) levels occurring in pregnant women [14]. On the other hand, the pregnant woman presents progressive neutrophilia during gestation which leads to a situation of major inflammation facilitating the work [2]. The role of hormonal factors could also be raised because of the induction of neutrophilic dermatoses such as Sweet's syndrome by oral contraceptives or their occurrence during pregnancy [15]. The treatment of the PSPG is not unique and is identical to that of the PG occurring *de novo*. It is based on the avoidance of debridement and systemic corticosteroids (0.5 to 1 mg/kg/day) [16,17]. However, prevention of PSPG in predisposed patients can be achieved by prophylactic perioperative corticosteroids in the case of planned surgery. In the case of emergency surgery, this treatment should be started immediately after surgery.

Conclusion

Our observation, as well as other cases of pregnancy-induced PG described in the literature, suggest that there is a link between pregnancy and PG. Pregnancy may be a triggering factor or promoter of the PG in the same way as, rheumatological, intestinal and haematological pathologies. In our observation, it seems that the pregnancy has triggered the recurrence of dermatosis, and that the caesarean has facilitated the clinical expression by the pathergy phenomenon.

Conflict of Interests

None.

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