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QATAR MEDICAL JOURNAL

CASE REPORT

Pyoderma gangrenosum following a routine caesarean section: Pseudo-infection in a caesarean wound

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http://dx.doi.org/10.5339/qmj.2015.1
Submitted: 29 October 2014
Accepted: 11 December 2014

Cite this article as: Radhika AG, Singal A, Radhakrishnan G, Singh S. Pyoderma gangrenosum following a routine caesarean section: Pseudo-infection in a caesarean wound, Qatar Medical Journal 2015:1 http://dx.doi.org/10.5339/qmj.2015.1

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ABSTRACT

A 22-year-old booked primigravida underwent lower segment caesarean section for breech presentation. She developed signs and symptoms of wound infection by the fourth postoperative day. This was initially managed with antibiotics and wound dressing, but debridement was later undertaken after consulting surgeons. This resulted in an alarming worsening of the wound with sudden and fast increase in its size along with systemic symptoms. Wound biopsy established the diagnosis of pyoderma gangrenosum. The patient's management included oral medication with prednisolone, cyclosporin and dapsone and wound care. There was a dramatic response to this treatment. The wound completely healed by the eighth postoperative month. The oral medications were tapered off slowly and stopped by that time.

Keywords: caesarean section, pyoderma gangrenosum, pathergy, non-healing ulcer

INTRODUCTION

Pyoderma gangrenosum (PG) was first described in 1930 by Brunsting et al.,[1] as an acute neutrophilic dermatosis. The epidemiology of PG has never been formally assessed in a population-based study.[2] Based on various case reports, case series and cohort studies, the incidence is believed to be about 3 to 10 cases per million per year,[3] though it has not been assesed in gynaecological patients. Females are genetically more predisposed than males. It is known to affect most commonly lower limbs however, any site can be affected. The typical clinical presentation is the development of an erythematous papule or pustule which rapidly progresses to an ulcer. This characteristic ulcer is painful with raised

violaceous borders. These lesions can develop spontaneously or following trauma and surgery.[4] Its aetiology is poorly understood. There are only a few reports of this condition following obstetric and gynaecological procedures. We share our experience of managing a case of pyoderma gangrenosum following caesarean section, which highlights that an easily misdiagnosed condition can result in extensive physical and psychological morbidity.

CASE REPORT

A 22-year-old, primigravida, underwent elective caesarean section for a term breech pregnancy. A healthy female baby was delivered weighing 2.6 kg. The antenatal period was uneventful. Routine post-operative care including broad spectrum antibiotics (third generation cephalosporins and antibiotics with anaerobe coverage) were given intravenously as per the institutional policy for 48 hours followed by oral medication. Recovery was satisfactory for the initial four days then, on the fifth postoperative day, the patient developed a fever of 38°c with increasing pain in the caesarean wound. On examination, there was mild abdominal distension with marked erythema and tenderness along the entire length of the caesarean wound extending for about 4 cm away from the incision line. Bowel sounds were present. Lochial discharge was healthy and there was no other apparent focus of infection. Ultrasound did not reveal any significant findings. Laboratory testing for neutrophilic leukocytosis was performed and cultures for blood, urine, wound site and vagina were also tested, keeping in mind resistant infection. Other tests included chest x-ray and widal tests for malarial antigens, all found to be normal.

Wound dressing was continued for a further two days and oral antibiotics were changed to broad-spectrum injectable antibiotics i.e., tazobactam and piperacillin in anticipation of resistant infection. All cultures reported on the seventh postoperative day were sterile confirming no growth of bacteria, fungi or tubercular organism, but there was worsening of the wound with purulent discharge and deepening erythema. The stitches were removed to release tension but this only resulted in the breakdown of the wound with a large area of ulceration. Consultation was sought from senior colleagues in the surgery department. A provisional diagnosis of deep-seated fulminant wound sepsis was made and debridement of the wound was performed. This only resulted in an alarming deterioration of the wound over the next two days. There was a dramatic increase in its size with development of raised pinkish-violaceous edges. By two weeks post-caesarean, there was a large area of raw ulceration extending to the umbilicus (Fig. 1). Considering the very unusual behaviour of the wound and the extensive flare-up after debridement, opinion was sought from a consultant dermatology who suggested a strong possibility of pyoderma gangrenosum with an urgent need for confirmation with biopsy from the wound margin. Histopathology report of the biopsy stated dermal neutrophilic abscess and an intense infiltration of granulocytes in the dermis with subdermal vascular occlusion suggesting pyoderma gangrenosum (Fig. 2). Other serologic tests, namely anti-phospholipid antibodies, rheumatoid factor and anti-double stranded DNA were carried out and all were negative. These tests were performed as pyoderma gangrenosum is known to have a strong association with inflammatory bowel disease, systemic lupus erythematosus (SLE), arthritis, etc.

In view of this unusual diagnosis, the case was reviewed again. The patient's history was revisited and physical examination repeated. A large scar was found on the right buttock. Her mother gave history of a huge ulcer having formed following an intramuscular injection on the buttock during early childhood. It had taken very long for it to heal and had left a deep scar at the injection site. There however were no ulcerations on any other part of the body including the lower limbs following minor trauma. She was treated by a multidisciplinary team including a dermatologist who helped in diagnosing the disease, an obstetrician for wound care and drug treatment and a psychiatrist for psychological counselling of the patient.

She was started on once daily doses of oral prednisolone 40 mg, dapsone 100 mg, cyclosporine 100 mg and analgesics (non-steroidal anti-inflammatory drugs and tramadol). Local treatment included biweekly dressing with normal saline and paraffin gauze. Within 48 hours of this treatment, there was dramatic improvement. She became afebrile; the erythema around the wound had waned followed by steady improvement in the following weeks. However, there were recurrent episodes of bacterial infections which were treated with appropriate antibiotics. To reduce superadded infections, biweekly dressings were started with silver nanocrystal dressings.

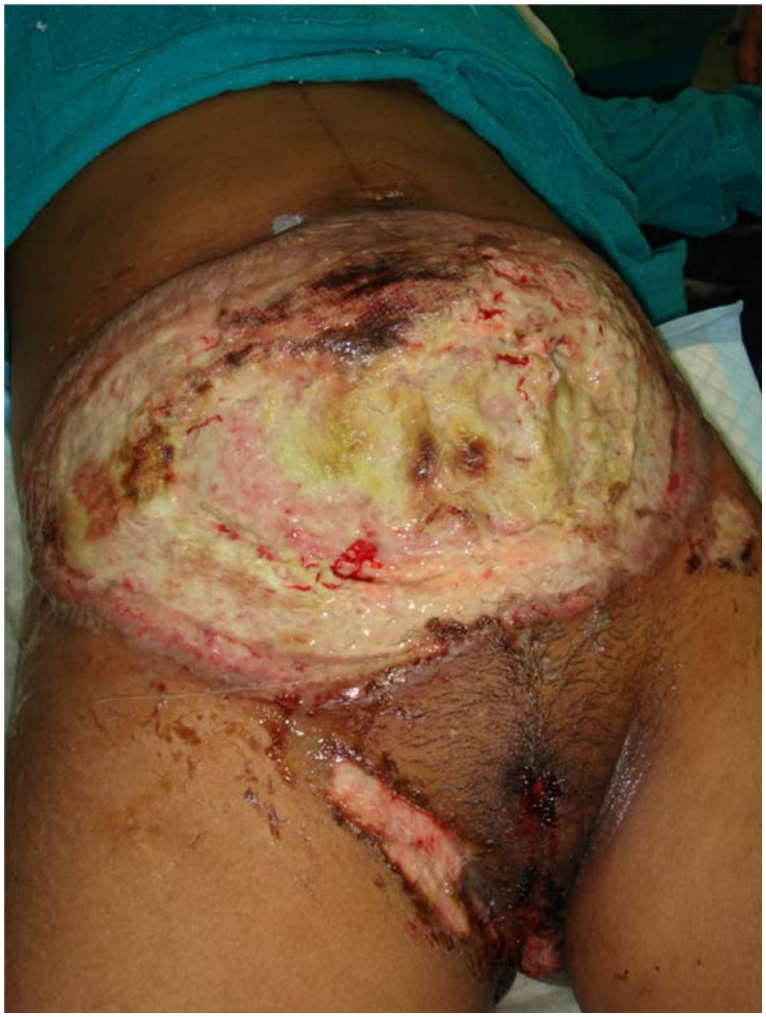

Figure 1. Large area of raw ulceration extending to the umbilicus from the incision site (two weeks post-caesarean).

This was continued for about four weeks and then followed up with 1% acetic acid covered paraffin gauze. Within about eight weeks of this treatment, there was growth of healthy granulation tissue and flattening of wound edges with disappearance of erythema.

Prednisolone was slowly tapered and stopped by four months. Dapsone and cyclosporine were continued together with biweekly dressings till complete wound healing, which was eight months post-caesarean, though, it had left a big scar (Fig. 3).

DISCUSSION

Pyoderma gangrenosum is a non-infectious neutrophilic dermatosis of unknown cause, often associated with other chronic inflammatory diseases such as arthritis, inflammatory bowel disease and haematological disorders in up to 50% of cases.[5] Its chief features include female predilection, frequently seen in adults between 20-40 years, more often involving the lower extremity. The lesions may develop spontaneously, after surgery or after minor trauma

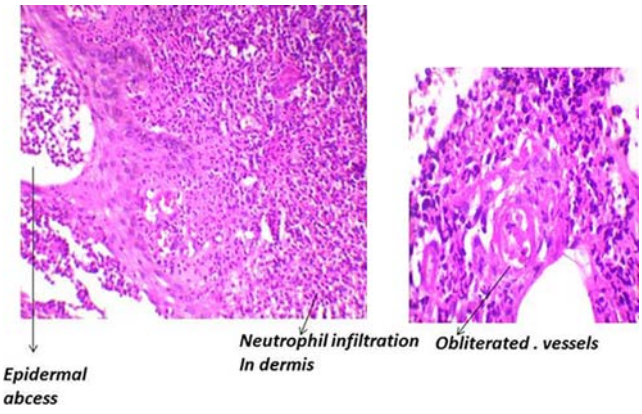

Figure 2. Histopathology slide showing acute neotrophilic dermatosis and intense infiltration of granulocytes in dermis.

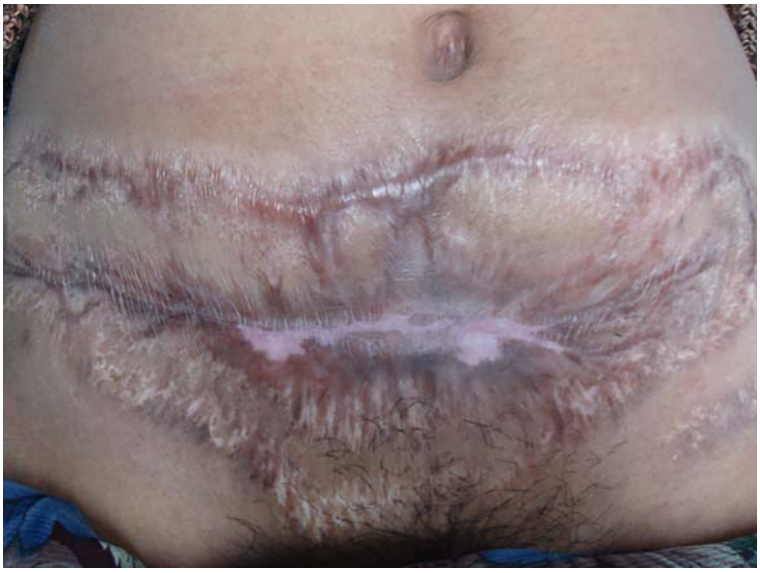

Figure 3. Scar site at eight months post-caesarean.

due to the pathergy phenomenon.[6] Pathergy is an exaggerated skin reaction to minor trauma. It is characterized by red bumps, pustules and/or ulceration. Its aetiology remains obscure. Various theories for its causation include dysregulation of the automimmune system,[7] especially aberrant neutrophilic migration[8] and genetic predisposition. There is a predisposition to pyoderma gangrenosum during pregnancy and postpartum.[9] Pregnancy is associated with immunosuppression, including IL-2 and IL-1 inhibition and depressed polymorphonuclear leukocyte (PMN) chemotaxis and adherence functions. It has been suggested that these alterations in the immune function of pregnant women could play a part in the development of PG in pregnancy.[10,11] About 50–70% of cases are associated with other systemic involvement such as ulcerative colitis and Crohn's disease, arthritis, haematological disorders (leukemia and monoclonal gammapathies), HIV infection, SLE and hepatitis.[6,12,2]

Pyoderma gangrenosum has been classified into four major clinical types: ulcerative; pustular; bullous; and vegetating or superficial granulomatous.[5] Its symptoms include severe wound site pain, fever and malaise, development of an erythematous papule or vesiculopustule, ulcer with violaceous colored borders and purulent discharge. Lesions can be single or multiple, chronic or recurrent. Surgical debridement worsens the condition due to a pathergic response.

Early diagnosis can be established by clinical features such as unusual flare-up of a wound following trauma, debridement or surgery; similar history of an extensive wound in the past or in family history; sterile wound with raised violaceous border and purulent discharge; histopathology supporting the diagnosis

Table 1. Pyoderma gangrenosum lesions post-surgery case reports in the literature.

	Author	Year	Day	Associated disease	Treatment
1.	Shand et al.[14]	1987	5 days	Family history	Steroid
2.	Harland et al.[15]	1993	5 days	None	Steroid + Surgery
3.	Stone et al.[16]	1996	Post cs	None	Steroid + Surgery
4.	Steadman et al.[17]	1998	1 day	Hypo-gammaglobulinemia	Steroid
5.	Ronnau et al.[18]	2000	6 days	Hepatitis C	Steroid + Cyclosporine
6.	Banga et al.[19]	2006	5 days	None	Steroid
7.	Sanz – munoz et al.[20]	2008	9 days		Steroid
8.	Pauser et al.[21]	2009	6 days	None	Steroid + Cyclosporine + Dapsone
9.	Our case	2013	3 days	None	Steroid + Cyclosporine + Dapsone

mainly by exclusion of other causes. Dermal neutrophilic abscess is the key histopathological feature, though nonspecific. Other causes of similar skin ulcerations include bacterial, fungal and viral infections, vasculitis, malignancy, vascular occlusive or venous disease and inflammatory disorders.[2]

PG has rarely been reported following surgery, but has been reported following laparoscopy in men[5] and following breast surgery in women.[13] There are also very few cases reported in women following delivery and caesarean section. Our review of the literature in Table 1 identified eight case reports with development of PG lesion post-surgery.

Moist wound management is a cornerstone of wound management. Topical corticosteroids are used to maintain remission.[22] Surgical debridement is contraindicated since it can be an aggravating factor for the disease as was seen in the present case.

The choice of treatment is prednisolone with cyclosporine and occasionally with dapsone to suppress neutrophil migration.[23]

ACKNOWLEDGEMENTS

We acknowledge the wholehearted support and hard work of many residents and staff nurses who were in contact with the patient at various times during the course of her stay in the hospital.

AUTHOR CONTRIBUTIONS

AGR was directly involved in management and treatment, wound dressing and drafting of the case report; AS was involved in diagnosis and advice on management; GR was involved in overall supervision and SS was involved in patient care and editing of the article.

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