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RARE CASE OF RECURRENT PYODERMA GANGRENOSUM IN PREGNANCY WITH EPISIOTOMY WOUND GAPE IN TERTIARY CARE HOSPITAL

Obstetrics & Gynaecology

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ABSTRACT

Pyoderma gangrenosum (PG) is a rare neutrophil-predominant inflammatory disease that initially presents as a sterile pustular lesion and may progress to painful non-healing ulcers with undermined violaceous borders. It is uncommon in pregnancy. As the root cause is unknown, the presentation is commonly associated with systemic inflammatory conditions such as inflammatory bowel disease, arthritis and hematological abnormalities. Diagnosis is difficult to make, mostly resting on the exclusion of other similar conditions.We describe a case of a postpartum patient with PG (as a rare case) who was diagnosed as having episiotomy gape and fever after normal vaginal delivery and was treated by teicoplanin, linezolid and steroids. Obstetricians need to understand the pathogenesis of PG, its clinical presentation, complications and the conditions associated with it because early diagnosis and proper targeted therapy will provide better outcome of patients in a tertiary care hospital.

KEYWORDS

Pyoderma Gangrenosum (PG), Pregnancy, steroids

INTRODUCTION

Pyoderma gangrenosum (PG) is a rare neutrophilic chronic inflammatory dermatosis that usually presents as a non-healing painful ulcer with violaceous borders.[1] It was first described by Brunsting et al [2] in 1930. Initially it was thought that lesions were produced by Streptococcus species and named the condition pyoderma gangrenosum, a purulent infection of cutis produced by pyogenic bacterial species.

Several systemic conditions like inflammatory bowel disease, arthritis, hematologic malignancies have been associated with the disorder.[3] It has predilection for the lower legs. The provocation of new lesions by trauma known as phenomenon of pathergy is seen in roughly 30% of patients with PG. [4] The diagnosis of PG is difficult and requires the exclusion of other causes of nonhealing ulcers like infections and neoplasms. Although still unclear, pathogenesis of PG is thought to be an immune-mediated process involving neutrophil dysfunction, abnormal inflammatory cytokines, and abnormal production of tumor necrosis factor (TNF)-α (a powerful proinflammatory cytokine). [5,6] Granulocyte macrophage colony-stimulating factor (GM-CSF) [11], a known attractant of neutrophilic inflammation is elevated in pregnancy and amplify the risk of neutrophil-driven PG in response to local trauma. Corticosteroids, azathioprine, mycophenolate mofetil, cyclophosphamide, infliximab, methotrexate, and intravenous immunoglobulin have been used in its systemic therapies. [4,7]

PG during pregnancy is quite uncommon. [8,9,10,11] Treatment of PG in pregnancy can be challenging. Therapy of pyoderma gangrenosum involves the multidisciplinary approach and use of local dressing, anti-inflammatory and immunosuppressive agents. Among systemic therapies, corticosteroids and cyclosporine were most commonly used to treat PG in pregnancy. [9] Intravenous immunoglobulin is another relatively safe option in pregnancy. [12] The prognosis of pyoderma gangrenosum is generally good; however, recurrences may occur and residual scarring is common.

CASE REPORT

27 years old G2P1L1 (Gravida 2, Para 1, Live 1) came to labour room, 9 months ANC with referral suggestive of sensitive to imipenem and meropenem. She had complaints of PV leak and pain in abdomen since 4 hours. The pain was in the lower abdomen radiating to back, increasing with time. She had no history suggestive of preeclampsia toxaemia or any other risk factors. She had married life for 10 years with 1st spontaneous conception 7 years back and this was her 2nd pregnancy. ANC was registered with proper follow-up and medications. Her gestational age was 39 weeks and 4 days by 1st trimester scan. She had past history of some skin lesions during her previous delivery but the details about the type of lesion and treatment were not available. Patient was in spontaneous labour, delivered female child and was discharged after 4 days uneventfully. Patient didn't have any skin lesion at the time of discharge.

On 6th day of discharge, patient presented with continuous fever of 101-degree F with no chills or diurnal variation and vesicular blebs with clear fluid with surrounding erythema, extremely painful, coming in crops all over the body since 2 days.

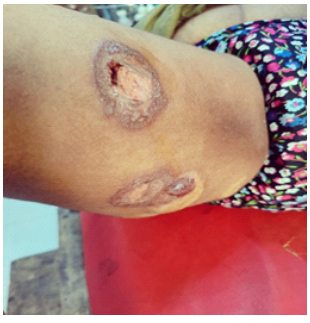

Figure 1 : Illustrating 2 lesions on right forearm

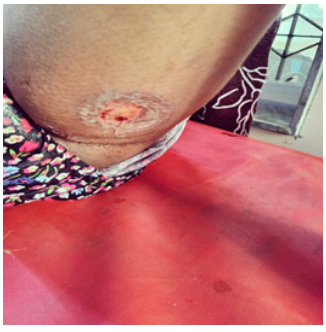

Figure 2 : shows one lesion on right thigh

On admission, patient was conscious, oriented, general condition - moderate, SPO2- 99% off O2, Pulse- 100/min, regular, low volume with BP- 120/70 mmHg, RR- 20cpm, Pallor +, CVS- S1 S2 heard, no murmur, RS- air entry bilaterally equal, no added sounds. Per abdomen examination- abdomen normal, 16 weeks uterus with involution. Local examination- episiotomy wound gape present with normal lochia. There were multiple tense fluid filled bullae over an erythematous base with tenderness present all over the body. Provisional diagnosis was P2L2 with post discharge day 6 with fever,

rash and episiotomy wound gape. She was started on T. Augmentin 625mg, T. Diclofenac 50mg and Inj Paracetamol twice daily. Fusiwal B cream application on the lesion locally. Investigations revealed haemoglobin- 9.2 g/dl, platelet- 1.5 lac /cmm, TLC- 6000/cmm, blood group-A positive, blood urea-23mg/dl, s. creat-0.9mg/dl, total bilirubin-0.7mg/dl, direct bilirubin-0.4mg/dl, indirect bilirubin-0.3mg/dl, SGOT-14IU/dl, SGPT-10IU/dl, RBS – 115g/dL, PT-20s, INR-1.43, CRP – 1.804 positive. All fever lab (PSMP, Dengue, Widal) were negative. Chest X-ray and USG A+P done. USG revealed involuting uterus. Skin lesion biopsy sent. Episiotomy wound dressing done twice daily. Her fever and rash didn't settle completely inspite of treatment. Patient had surge of fever on 4th day of re-admission of 101- and 102-degree F, vitally stable with haemoglobin- 7.5g/dL, WBC – 24000/cmm, platelet – 2.4 lac/cmm, LFT, RFT, ANA, dsDNA were normal. Diagnosis was still not confirmed as skin biopsy and blood culture report were awaited. On the basis of lesions, provisional diagnosis of ? pyoderma gangrenosum was acted upon by starting patient on Inj Teicoplanin 400mg i.v. loading dose followed by maintenance dose of 200mg i.v. and T. Linezolid 600mg for 10days twice daily empirically. Her fever subsided after 8th day of re-admission but her skin lesions didn't resolve. Every 3rd day her WBC was repeated and it had decreasing trend from 24000 to 18000 to 11000/cmm on 10th re-admission day. Biopsy report was s/o pyoderma gangrenosum. When WBC count was 11000/cmm, inj Methylprednisolone 500mg i.v. OD was started for 3 days followed by T. prednisolone 48mg OD for first 7 days, 24mg for next 7 days, 12mg-8mg-4mg for successive 7 days for complete 1 month course of steroid. Wound dressing with betadine and Dermadue cream (for hydration) was done every 3rd day. Lesion started to resolve on calf and upper limbs but thigh lesions were still in healing phase. Episiotomy re-suturing done on day 13th of re-admission. She was discharged after 15 days on T. prednisolone. She followed up later with complete remission of skin lesions.

DISCUSSION

Pyoderma gangrenosum belongs to one of the rarest non-infectious neutrophilic dermatoses. Many patients fall in the age group of 20 to 50 years with women being affected more than men. Lesions can develop spontaneously, after surgery or after minor trauma (pathergy phenomenon). They can be single or multiple, chronic or recurrent. It presents as severe wound pain, fever, and malaise. There is development of an erythematous papule or pustule initially that breaks down to form an ulcer with purulent discharge as well as violaceous coloured borders with undermined edges. The early stages may resemble a bacterial infection and diagnosis is made by exclusion of other cutaneous conditions. Histopathology of biopsy taken from ulcer edge shows a deep suppurative folliculitis with dense neutrophilic infiltrate.

Here in our case, patient was treated with teicoplanin and linezolid first due to her prior history of MRSA detected from similar skin lesions and raised WBC count. As soon as her WBC count came to normal, she was started on systemic corticosteroids which is the first line of treatment for PG. Treatment includes topical therapies like local wound care and dressings with betadine and hydration maintaining agents. Drastic improvement with corticosteroid treatment supports the diagnosis of pyoderma gangrenosum. Prednisolone,1 to 2mg per kg per day, is widely used as initial therapy [13]. Dapsone (diaminodiphenylsulfone) in the doses of 100 to 200mg per day in divided doses and cyclosporin are another popular drugs that have been accepted as treatment for widespread PG after initial steroids or in combination with steroids. Those patients for whom steroids cannot be given for some reason, Azathioprine (100 to 150mg/day) can be given with latency period of 2-4 weeks. Recently infliximab has been shown to be effective in a single dose of 5mg/kg given in day, thereafter repeated at 4 to 6 weeks intervals. Intralesional cyclosporine, azathioprine, chlorambucil, mycophenolate and several other drugs have been used with various success. Autologous split skin graft can be used in extensive skin tissue loss. 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 and not available in all the institutions.

CONCLUSION

We weren't fortunate enough because diagnosis and treatment were not straight forward in this case as patient neither gave complete past history nor had previous reports or records. Proper treatment was possible after excluding the differential diagnosis. PG is rarely encountered during pregnancy. Most of the cases reported during pregnancy and puerperium have coexisting disease conditions, while our case had none. Early diagnosis and subsequent treatment are crucial for limiting scar tissue. Although it is a dermatological condition, an obstetrician must know how to diagnose it early to decrease the complication and discomfort of the patient.

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