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Pyoderma Gangrenosum after Minimally Invasive Cardiac Surgery: A Nightmare Experience

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Abstract

Pyoderma Gangrenosum (PG) is an autoinflammatory condition mimicking Surgical Site Infection in the postoperative period and both these conditions differ entirely in terms of their management. Early and prompt diagnosis is crucial to reducing surgical morbidity. We present a postoperative case of Ostium Secundum Atrial Septal Defect (OS ASD) closure using a minimally invasive approach from right sided Trans Axillary site who developed an ulcerative form of PG at the surgical site and how timely diagnosis guided us in managing the patient with successful outcomes.

Keywords

Postoperative Pyoderma Gangrenosum, Minimally Invasive Cardiac Surgery, Ostium Secundum Atrial Septal Defect, Methyl Prednisolone, Mycophenolate Mofetil

1. Introduction

Pyoderma Gangrenosum is a very rare autoinflammatory cutaneous disorder with unknown etiology and mostly associated with systemic inflammatory diseases. Operative procedure or trauma are provoking factors known as pathergy. Till date very few cases are documented post cardiac surgery mostly after coronary artery bypass grafting. Pyoderma Gangrenosum is a diagnosis of exclusion which starts as pustules or bullae that progress very rapidly as deep ulcers with ragged margins and undermined edges. We report a case of young 26 years old female patient who

underwent Minimally Invasive Atrial Septal Defect Closure (ASD) via a right trans axillary approach diagnosed with PG.

2. Case Report

26-years-old female presented to the OPD with complaints of dyspnoea on exertion and chest pain since one year for which she was evaluated and diagnosed with large Ostium Secundum ASD (OS ASD) for which, she underwent Minimally Invasive Dacron patch closure of OS ASD via right trans axillary approach through central cannulation. A vertical midaxillary skin incision of (approximately 5 cm length) was given and the pleural cavity was entered through the fourth intercostal space and Dacron patch ASD closure was done. Ceftriaxone was given as part of the perioperative antibiotic course. She had an uneventful hospital stay and was discharged on 3rd postoperative day. On 7th postoperative day, the patient came for follow up presenting with complaints of painful wound with redness and pus discharge from surgical site of 2 days duration for which she was admitted & started on intravenous antibiotics. In view of suspecting infection, blood, urine and wound cultures, were sent followed by regular debridement and dressings for a week. Wound culture report was negative for bacterial growth and wound progressed to deep ulcers of 10 × 10 cm with an erythematous to violaceous border and area of necrosis, everted hard edges along with 3 separate ulcerative lesions of 1 cm each around the main ulcer with engorged breast and mild retraction of nipple with 3 × 3 cm of firm palpable mobile central axillary node, progressing to right upper chest and back (**Figure 1**, **Figure 2**). Since the time of re admission, multiple wound cultures were sent and various antibiotic therapy regimens were administered such as injectable Linezolid and Clindamycin twice daily for five days, Ceftriaxone 1 gm twice a day and Meropenem 1 gm thrice a day for two weeks, Gentamycin 80 mg twice daily for 10 days. The patient's investigation results at that time of readmission are presented in **Table 1**.

Table 1. Laboratory values on readmission.

Labs	Patient value	Reference range
1) White blood cells (WBC)	11.27 × 10^3/mcL	4.5 - 11.0 × 10^3/mcL
2) PROLACTIN	46.26 ng/ml	3.8 - 23 ng/ml
3) C-reactive protein	331.91 mg/L	<9.9 mg/L

During serial debridements, the WBC counts raised to 19.79 × 10^9 cells/L and later to 31 × 10^9 cells/L. Despite all the above treatment measures, the wound did not show any signs of healing following which an alternative diagnosis of PG was considered following consultation with both dermatology and general surgery departments and patient was started on Dexamethasone 8 mg once a day for 5 days and later on changed to Injection Methyl Prednisolone 40 mg once daily for 10 days which was later tapered gradually over 4 weeks. The wound margins showed signs of healing with healthy granulation tissue only after initiating steroid ther-

apy. The wound was dressed with amorphous hydrogel and colloidal silver ointment (Hydrogel AM, Eris Oaknet Healthcare Pvt Ltd.) and normal saline soaked gauze. Mycophenolate mofetil (MMF) 500 mg once a day for a week was started after steroids were tapered off and then dosage was escalated to twice a day after Pulmonology clearance. Meanwhile other factors such as anemia and hypo albuminia were corrected with blood transfusions and high protein diet. After 40 days when the wound was healthy (Figure 3), collagen sheet dressing with normal saline was done and paraffin gauze was applied over it. Patient was discharged with stable hemodynamics and on oral Methyl Prednisolone 20 mg once a day and MMF 1000 mg in morning and 500 mg during night along with instructions regarding dressings and wound care management while at home. Improvement in the wound was noted within one week of starting this treatment (Figure 4).

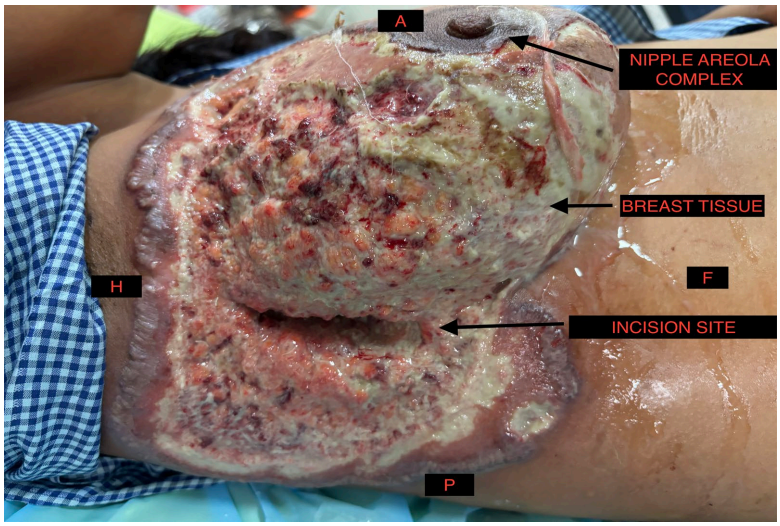

Figure 1. Photograph shows original midaxillary incision site and extensive involvement of wound and entire breast tissue except nipple areola complex and typical necrotic tissue with ragged undermined margins with pustules at border (H—Head end, F—Foot end, A—Anterior, P—Posterior).

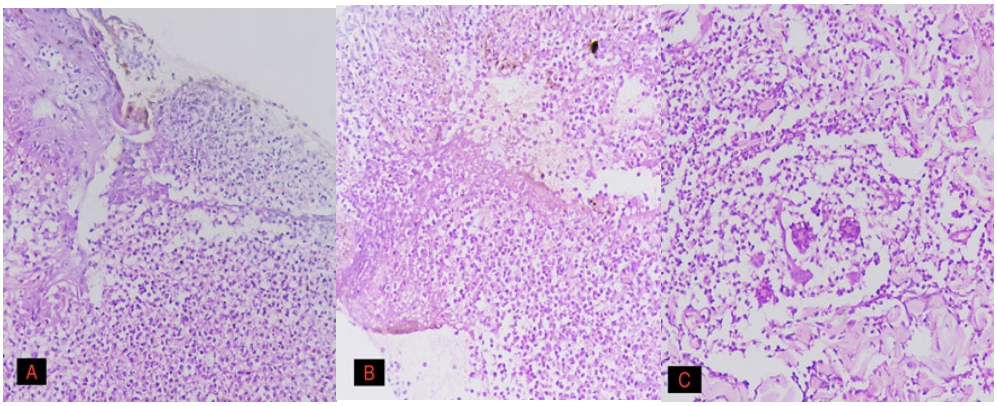

Figure 2. Histopathological images of biopsied tissue margins. (A) Superficial epidermal necrosis with dense neutrophilic infiltrate; (B) Leukocytoclastic vasculitis; (C) multinucleate giant cell.

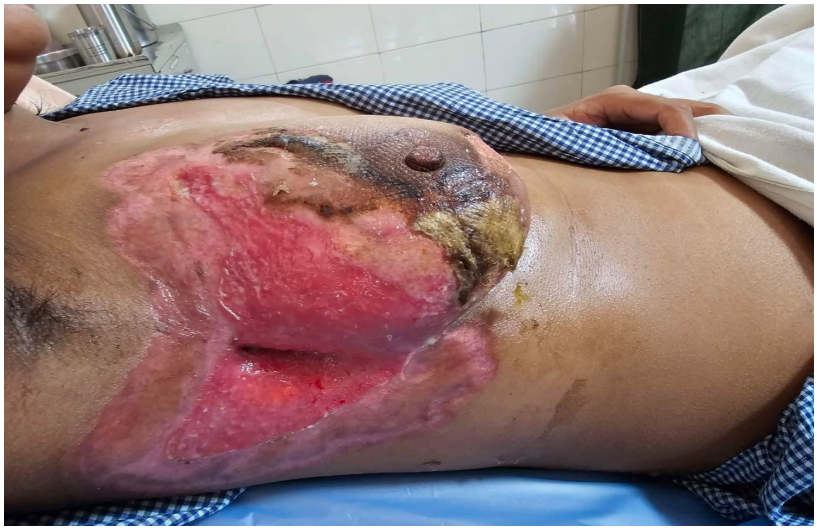

Figure 3. Surgical site 4 weeks after initiation of steroid therapy.

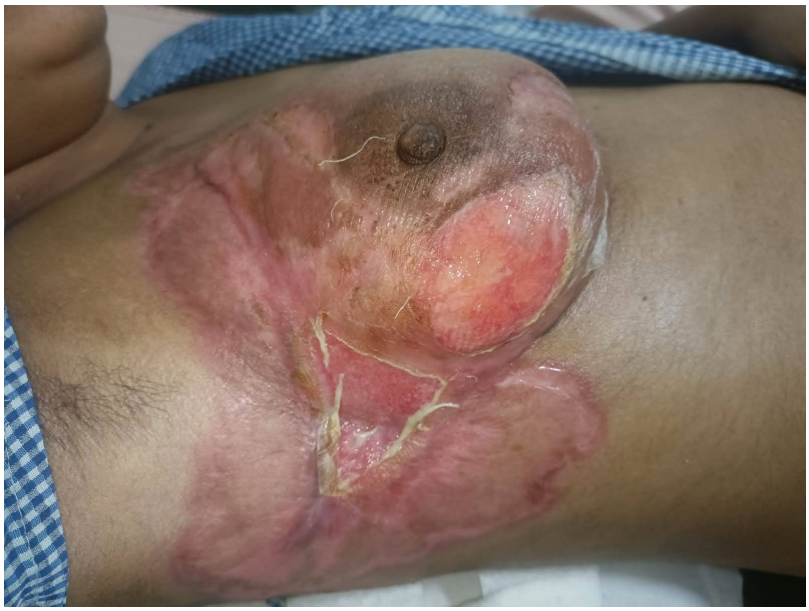

Figure 4. Surgical site post collagen sheet dressing and 6 weeks after initiation of Steroid therapy.

3. Discussion

Postoperative PG (PPG) is a skin disorder and a diagnosis of exclusion associated with systemic illness such as haematological dyscrasias being the most common in 50% - 78% cases [1]-[3]. Breast and abdomen are the most common sites occurring at approximately 7 days after the surgery [4].

PPG is characterized by fast progressive painful necrotic ulcers involving all layers of the skin. PPG is divided into four subtypes: 1) Pustular; 2) Vegetative; 3) Bullous; and 4) Ulcerative. Haematological disorders are mostly associated with bullous PG and Pustular PG is usually associated with inflammatory bowel disease. Other rare variants of the PG are vesiculo pustular, extracutaneous variants

and malignant pyoderma [5]. The present case is classified as Ulcerative subtype of PPG. Many evidences suggest immunological response to unknown etiology for PG. Tumor Necrosis Factor (TNF) and Interleukin 1Beta (IL1-B) are most commonly associated cytokines with PG and various medications used in the treatment of PG targets these cytokines [6].

In the existing literature, Post-operative PG has been reported commonly after abdominal and thoracic surgeries and rarely reported after Coronary Artery Bypass Grafting (CABG) surgery [7]. Very few cases have been reported after Mitral valve replacement [8] and aortic valve replacement surgeries [9]. So far there are no published case reports of PPG post Minimally Invasive Cardiac Surgery. Diagnosis of PPG is not very common in the initial postoperative period because it mimics Surgical Site Infection (SSI) as noted in the present case despite the elevated serum Procalcitonin levels which is an incidental finding. PPG is considered as differential diagnosis when poor wound healing is noticed along with development of rapidly evolving ulcers having violaceous undermined borders with negative culture sensitivity reports and there is unresponsiveness to antibiotic therapy especially in female sex and in association with some systemic diseases as mentioned earlier mainly in patients undergoing breast or abdominal surgeries. In the present case there was no evidence of associated systemic illnesses such as hematological abnormalities since the Complete Blood Count and peripheral smear examinations were normal except leukocytosis with Neutrophilia and patient did not have any bowel related symptoms. Non-specific markers such as leucocytosis and elevated levels of C-Reactive Protein which are common for both SSI and PPG adds to the diagnostic dilemma and was the reason for the initial mis diagnosis as SSI in this case. Prompt diagnosis and starting of appropriate treatment in the form of systemic corticosteroids and immunomodulators is very crucial because delayed diagnosis increases morbidity and debridement should be avoided altogether as it induces a pathergy response which exacerbates PG lesions resulting in rapid wound deterioration as noticed in this case [4].

4. Conclusion

PG is a very rare complication after cardiac surgery and its management is diametrically opposed to that for its main differential diagnosis, Surgical Site Infection (SSI), which requires antibiotics and debridement. Early diagnosis and aggressive treatment with a multidisciplinary team approach involving specialists from Dermatology, Rheumatology, etc., is very crucial to avoid mismanagement for non-healing postoperative wounds that are culture-negative and unresponsive to antibiotics. It is also crucial to treat prophylactically with corticosteroids before any surgical procedures who has previously been diagnosed or treated for Pyoderma Gangrenosum.

Ethics Approval

The study has been conducted after approval from Institutional Ethics Committee

of All India Institute of Medical Sciences, Raipur, India, for reviewing the medical records of the patients bearing approval number IECSG-189/08-08-2023. The patient data and other information were used in compliance with the Declaration of Helsinki.

Informed Consent

Informed Consent has been obtained from the patient(s) to publish their information anonymously for academic purposes.

Conflicts of Interest

The authors declare no conflicts of Interest.

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