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Postoperative Pyoderma Gangrenosum: A Diagnostic Challenge

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1. Abstract

1.1. Background

Pyoderma gangrenosum (PG) is an autoinflammatory phenomenon of the skin, frequently linked with common autoimmune conditions such as rheumatoid arthritis and inflammatory bowel diseases. Postoperative PG (PPG) is an uncommon pathergy variant that often mimics surgical site infection, leading to repeated debridement's, inappropriate antimicrobial therapy, and delays in diagnosis.

1.2. Methods

We report the case of a 76-year-old woman with long-standing rheumatoid arthritis on immunomodulatory therapy who developed a painful ulcerative lesion at the site of multiple thoracic spinal surgeries with hardware implantation.

1.3. Results

Her postoperative course was complicated by recurrent wound dehiscence, vertebral osteomyelitis, and culture-proven infections with methicillin-sensitive Staphylococcus aureus (MSSA), extended-spectrum beta-lactamase (ESBL) producing Klebsiella pneumoniae, and Proteus mirabilis. Despite prolonged antibiotic therapy and repeated operative interventions, the wound continued to progress. Histopathology demonstrated a plasma cell–predominant infiltrate without classic neutrophilic features, and immunofluorescence was inconclusive. Given her prior history of PG on the leg, the clinical appearance of the wound, and the response to intralesional as well as topical corticosteroids, a diagnosis of PPG was favoured after multidisciplinary review. In the context of the patient's chronic osteomyelitis, systemic steroids were not considered an option for treatment.

1.4. Conclusion

Clinical suspicion, along with involvement of multiple disciplines, is essential in the timely and appropriate management of PPG. Through this case, we aim to bring about more awareness about the occurrence of a PPG and guide diagnosis, respectively

2. Introduction

Pyoderma gangrenosum is a rare skin disease that can occur in isolation or in association with autoimmune conditions such as inflammatory bowel disease or rheumatoid arthritis (RA). The association of pyoderma gangrenosum with rheumatoid arthritis is well known [1]. About half of the cases of PG are associated with a systemic condition [2]. PG is seen in females more than in males. The median age group for the occurrence of PG is from adolescence to the middle-aged groups [3]. The disease mostly occurs as an isolated phenomenon in patients with no underlying systemic conditions. PPG is most described on the chest, abdominal wall, and procedures involving the breasts [4]. Development of PPG is thought to be secondary to pathergy. Pathergy is a phenomenon where minor trauma to the skin can lead to hyper-reactive skin lesion such as PG. In PPG, the inciting event is the skin incision which then further worsens the lesion with repeated interventions such wound debridement [5]. The incidence of PPG is rare, with fewer than 100 postoperative cases reported overall, and only a small subset occurring after spinal surgery.

3. Case Presentation

We describe a case of a 76-year-old female patient with advanced rheumatoid arthritis on long-standing immunomodulating therapy who developed a PG on her dorsal vertebral skin after multiple spinal surgeries. The patient initially had multilevel thoracic vertebral compression fractures for which she underwent corpectomy and hardware implantation.

The patient then developed vertebral osteomyelitis from an enlarging phlegmon, which was complicated by severe cord compression due to bone trauma. The patient's history included rheumatoid arthritis treated with hydroxychloroquine 300 mg daily and prednisone five mg daily. The clinical course was complicated by repeated incision and drainage after developing wound dehiscence. The wound progressed into a dendritic, branching

lesion with violaceous borders that did not respond to chronic antibiotic therapy or robust wound care Figure 1.

The patient had confounding microbiological data wherein first MSSA was identified and then ESBL Klebsiella pneumoniae and multi sensitive Proteus mirabilis at the wound site in the following months. The patient received about 5 weeks of intravenous ertapenem therapy followed by chronic dapsone and cephalexin therapy. The patient had multiple hospitalizations for wound dehiscence, spinal manipulation, hardware removal, and reimplantation. Wound evolution and staging suggested a concern for an underlying process not responding to chronic antibiotic suppression and dedicated wound care.

It is important to note that the patient also developed a PG on her left anterior leg a few years before this presentation. At the time, this was also presented in an atypical fashion and was misdiagnosed. The patient was on chronic immunosuppression at the time, and her leg lesion was treated with topical steroids. The diagnosis of PG was established after the involvement of multiple specialists, including dermatology, rheumatology, and infectious disease. The patient's lesion improved after periodic intralesional triamcinolone injections as well as daily clobetasol application. The patient was continued on dapsone as an adjunctive measure for PG. The patient's skin biopsy and histopathology did not reveal a neutrophilic infiltrate but did show a plasma cell infiltrate. Immunofluorescent staining was inconclusive.

Additional systemic immunosuppression therapy could not be a treatment modality for the patient due to the history of vertebral osteomyelitis and chronic kidney disease contraindicated cyclosporin therapy. The topical clobetasol and intralesional triamcinolone, however, brought about a substantial improvement in the wound Figure 1.

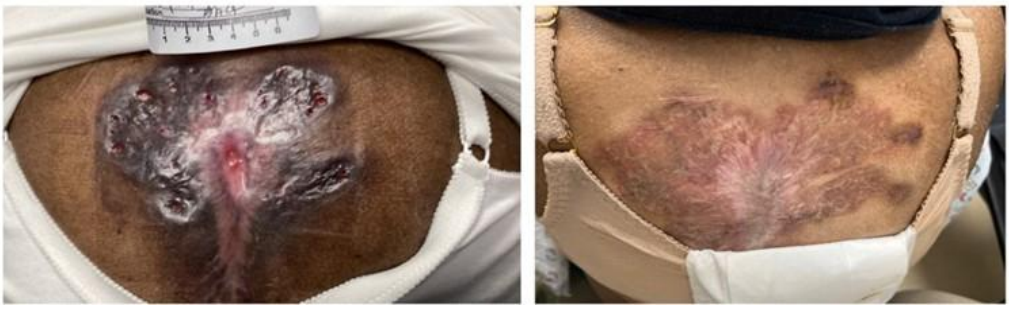

Figure 1: Pyoderma gangrenosum wound seen before and after 3 months of topical steroid therapy and oral dapsone therapy.

4. Discussion

The patient's pathology reports were not characteristic of neutrophilic dermatosis; however, with a trial of topical clobetasol, the wound started to recede. The diagnosis of PPG was clinical using the PARACELSUS score [6]. PPG often presents within days to weeks following surgery and demonstrates pain out of proportion, characteristic features of the violaceous, undermined borders and rapid ulceration. In our case, this led to repeated broad-spectrum antibiotic regimens and repeated debridement, which worsened the lesion due to pathergy.

Current literature suggests that if systemic immunosuppressive therapy with corticosteroids or cyclosporin is contraindicated for moderate to severe disease, then topical immunosuppression will be the modality of choice [7]. Systemic corticosteroids were deferred due to the patient's ongoing vertebral osteomyelitis. Given the rarity of PG at spinal surgical sites, our case contributes to the limited literature and aims to reinforce the principle that chronic post-surgical wounds, particularly in patients with autoimmune diseases, should prompt consideration of PG in the differential diagnosis.

5. Conclusion

Post-operative pyoderma gangrenosum is a rare but important diagnosis to consider in patients with chronic, non-healing surgical wounds, especially those with autoimmune conditions like rheumatoid arthritis. This case highlights the risk of pathergy from repeated surgical interventions and the importance of early clinical suspicion.

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