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Pyoderma Gangrenosum - A Case Report on Surgical Management

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Abstract: ***Pyoderma gangrenosum is a rare dermatosis of unknown etiology and variable clinical presentation. Pathogenesis includes, neutrophil dysfunction, immune dysregulation and abnormal cytokine signalling by T cells and macrophages. This is the case of a 28 year old female diagnosed with pyoderma gangrenosum. Patient was referred from department of surgery in view of progressive non healing ulcers on dorsum of both feet. A skin biopsy performed from the edge ulcer showed features suggestive of pyoderma gangrenosum with dense dermo-epidermal neutrophilic infiltrate and perivascular lymphocytic infiltrate. Patient received immunosuppressive therapy for 1 month following which she underwent skin grafting. This report highlights the possibility of combining adequate immunosuppressive therapy with skin grafting for a better aesthetic outcome in these patients.***

Keywords: Non healing ulcer, immunosuppression, split skin grafting

1. Introduction

Pyoderma gangrenosum (PG) also described by Brocq and Simon in 1908 as "*phagédénisme géométrique* is a rare non-infectious neutrophilic dermatosis commonly associated with underlying systemic disease. It has a worldwide estimated incidence of 3-10 cases/million people/year[1]. The incidence of PG increases with age, with a median age of 50 years. It is commoner in females with no association with ethnicity. The pathogenesis of pyoderma gangrenosum is not fully understood. It involves genetic mutations, neutrophil dysfunction, and immune dysregulation, abnormal cytokine signalling by T cells and macrophages. Lesions of pyoderma gangrenosum have been found to have increased levels of inflammatory mediators[2]

Clinically it presents with rapid progression of a painful necrolytic cutaneous ulcer with an irregular, violaceous and undermined border. Patients with pyoderma gangrenosum exhibit pathergy, a phenomenon whereby skin trauma provokes lesions at the site of injury.

The lower legs are most frequently affected although PG can present at anybody site. Subtypes of PG include bullous, vegetative, pustular, peristomal and superficial granulomatous variants.3

The various treatment modalities for PG include topical corticosteroids or calcineurin inhibitors in combination with systemic corticosteroids, cyclosporine A, azathioprine, cyclophosphamide, mycophenolate mofetil, intravenous immunoglobulins or monoclonal antibodies against TNFα. Surgical therapy is difficult due to risk of pathergy. Nevertheless, after having stopped the inflammation, the ulcers can be treated by split thickness skin grafts and simultaneous immunosuppression.4

The following case report highlights acceptance of skin grafting in a patient diagnosed with pyoderma gangrenosum following adequate immunosuppression

2. Case Report

A 28 year old female was referred from department of surgery in view of progressive non healing ulcers on dorsum of both feet since two months with worsening of ulcer despite regular dressings and appropriate wound care. It started as small elevated lesions which spontaneously evolved into rapidly progressive painful ulcers. All other causes for ulcers were ruled out. On examination - there were two well defined ulcers with irregular margins and peripheral violaceous rim on dorsum of both feet measuring roughly 5.5 x 7.5cms, floor contained slough and pale granulation tissue. Ulcers were tender and indurated on palpation, immobile and fixed to surrounding muscle and bone.

A skin biopsy performed from the edge ulcer showed features suggestive of pyoderma gangrenosum with dense dermo-epidermal neutrophilic infiltrate and perivascular lymphocytic infiltrate. Patient was started on tablet methylprednisolone 20mg in tapering doses for 1 month along with capsule minocycline 65 mg once daily for a month. Patient also had regular dressings and appropriate wound management following 1 month of immunosuppressive treatment she underwent skin grafting. Patient was reviewed 10 days, 16 weeks, 24 weeks post grafting. Patient showed signs of graft acceptance and good healing.

3. Discussion

PG is a clinically challenging frequently misdiagnosed condition with multiple mimickers like infections, vascular insufficiency, tissue injury etc. There are various treatment modalities available for pyoderma gangrenosum.

Debridement of the ulcers should be done gently using Burrow′s solution, silver nitrate or potassium permanganate. Topical and intralesional corticosteroids, topical 5-aminosalicylic acid, benzoyl peroxide, topical sodium cromoglycate, intralesional cyclosporine and topical nitrogen mustard can be used. Intralesional corticosteroid injections with triamcinolone acetonide may halt progression of ulcer and induce healing. 10% of 5-aminosalicylic acid suppresses leukocyte motility and cytotoxicity and helps in ulcer regression

Systemic corticosteroids are considered as the drug of choice for the treatment of PG, doses of prednisolone range from 40-80 mg/day. Dapsone in a dose of 100-200 mg/day, Sulfasalazine, sulfapyridine and sulfamethoxy-pyridazine have been shown to be effective in PG. The beneficial effect is by their ability to inhibit neutrophil chemotaxis

Minocycline in a dosage of 200-300 mg/day is beneficial in PG by its anti-inflammatory effect and by diminishing the chemotacticresponsiveness of neutrophils. Immunosuppressive agents like azathioprine (100-150 mg/day), mercaptopurine, cyclophosphamide (100-150 mg/day), arabinoside, chlorambucil, colchicine and daunorubicin have been used as adjunctive or alternative therapy to systemic corticosteroids with varying success in PG

Plasma exchange, intravenous immunoglobulin, hyperbaric oxygen therapy, thalidomide, nicotine, and potassium iodide may also been used[5]

Wound care and pain control are key features in the treatment of pyoderma gangrenosum Other therapies that have been successful are anti-TNF-alpha drugs such as etanercept and adalimumab.6

The ulcers of PG without skin grafting require a prolonged time to heal, being prone to secondary infection, which potentially represents an additional trigger for pathergy. In addition, long-term systemic immunosuppressive therapy is associated with adverse reactions necessitating other treatment modalities[7]. Recent advances have suggested a surgical approach of PG with split thickness skin grafting (STSG) as a safe and valuable treatment if performed under adequate immunosuppression. A case report by Ilknur Altunay et al demonstrated a case of colorectal adenocarcinoma with PG, who responded partially to topical treatments and systemic immunosuppressants and healed completely with surgical wound repair and hyperbaric oxygen therapy. Patient was given tablet prednisolone and cyclosporine for a month prior surgery following which they noticed complete healing and excellent aesthetic outcome[8]

A study done by Cliff, S et al demonstrated that split skin grafts were a useful treatment modality in 4 patients with ulcerative PG, producing a good cosmetic result. One case illustrated the importance of ensuring the disease is quiescent prior to grafting, to avoid pathergy. The other cases emphasised the need for prolonged immunosuppressive therapy to minimise the chance of reactivation of the disease process. They also concluded that the ultimate cosmetic result was superior after immunosuppression[9]

Another study evaluating efficacy of skin grafting in pyoderma gangrenosum in 153 patients found complete healing of wounds in 75.5% patients. The average time to complete healing was 10.8 weeks, mean donor site healing time was 1.9 weeks. Pathergy was reported in 8 (5.2%) patients

A statistically significant difference in the number of patients receiving preoperative and postoperative immunosuppressive therapy was found between the groups with complete healing/reduction and no improvement/aggravation[10]

While surgical treatment is supported by the published data, the exact dose and duration of required immunosuppression is still evolving.

4. Conclusion

In the above reported case, good surgical outcome was observed post surgery after an immunosuppressive therapy for 1 month. To limit the risk of pathergy developing, a role for prolonged courses of immunosuppressive therapy is suggested in previous literature. The most effective dose and duration of immunosuppressive therapy in patients with PG treated with split skin grafts remains to be determined

References

[1] Maronese CA, Pimentel MA, Li MM, Genovese G, Ortega-Loayza AG, Marzano AV. Pyoderma Gangrenosum: An Updated Literature Review on Established and Emerging Pharmacological Treatments. Am J Clin Dermatol. 2022 Sep; 23 (5): 615-634

[2] Shahid S, Myszor M, De Silva A. Pyoderma gangrenosum as a first presentation of inflammatory bowel disease. BMJ Case Rep. 2014 Nov 9; 2014: bcr2014204853. doi: 10.1136/bcr-2014-204853. PMID: 25385558; PMCID: PMC4225318.

[3] George C, Deroide F, Rustin M. Pyoderma gangrenosum-a guide to diagnosis and management . Clin Med (Lond). 2019 May; 19 (3): 224-228.

[4] Sick I, Trautner B, Ruzicka T. Chirurgische Therapie beim Pyoderma gangraenosum [Surgical management of Pyoderma gangrenosum]. Hautarzt. 2012 Jul; 63 (7): 577-83. German. doi: 10.1007/s00105-011-2269-8. PMID: 22146897

[5] Bhat RM. Management of pyoderma gangrenosum – An update. Indian J Dermatol Venereol Leprol 2004; 70: 329-335

[6] Pichler M, Larcher L, Holzer M, Exler G, Thuile T, Gatscher B, Tappeiner L, Deluca J, Carriere C, Nguyen VA, Moosbrugger-Martinz V, Schmuth M,

Klein GF, Eisendle K. Surgical treatment of pyoderma gangrenosum with negative pressure wound therapy and split thickness skin grafting under adequate immunosuppression is a valuable treatment option: Case series of 15 patients. J Am Acad Dermatol. 2016 Apr; 74 (4): 760-5.

[7] Eisendle K, Thuile T, Deluca J, Pichler M. Surgical Treatment of Pyoderma Gangrenosum with Negative Pressure Wound Therapy and Skin Grafting, Including Xenografts: Personal Experience and Comprehensive Review on 161 Cases. Adv Wound Care (New Rochelle). 2020 Jul; 9 (7): 405-425

[8] Altunay I, Kucukunal A, Sarikaya S, Tukenmez Demirci G. A favourable response to surgical intervention and hyperbaric oxygen therapy in pyoderma gangrenosum. Int Wound J. 2014 Aug; 11 (4): 350-3

[9] Cliff, S.1; Holden, C. A.1; Thomas, P. R. S.2; Marsden, R. A.3; Harland, C. C.1. Split Skin Grafts in the Treatment of Pyoderma Gangrenosum: A Report of Four Cases. Dermatologic Surgery 25 (4): p 299-302, April 1999

[10] Morgenstjerne-Schwenck LET, Knudsen JT, Prasad SC. Efficacy and safety of skin grafting in treatment of vasculitic ulcer and pyoderma gangrenosum-A systematic review. Wound Repair Regen. 2021 Mar; 29 (2): 240-253

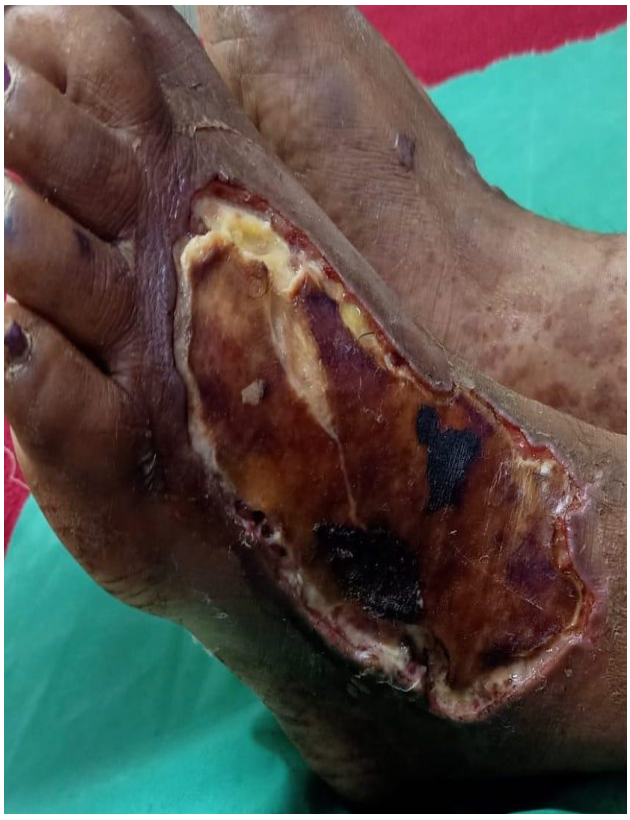

Figure 1

Figure 1: At Presentation - well defined ulcers with irregular margins and peripheral violaceous rim on dorsum of both feet measuring roughly 5.5 x 7.5cms. Floor contained slough and pale granulation tissue

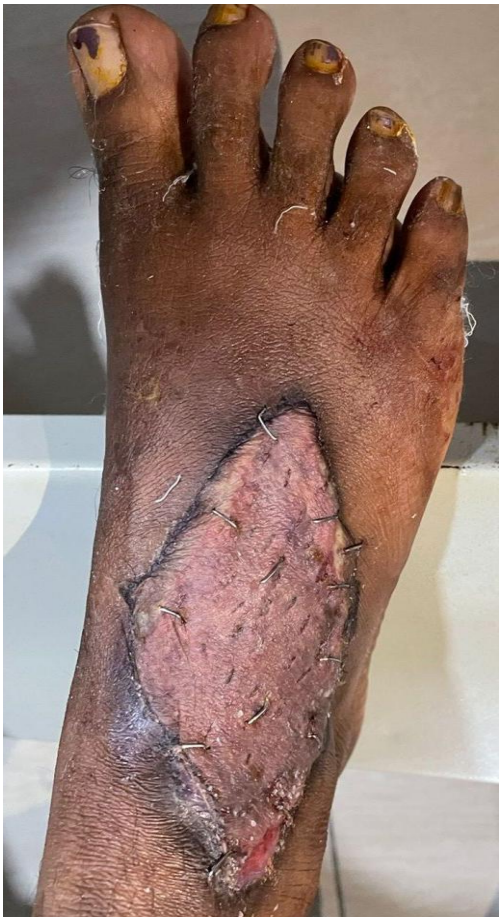

Figure 2: 10 Days Post Grafting

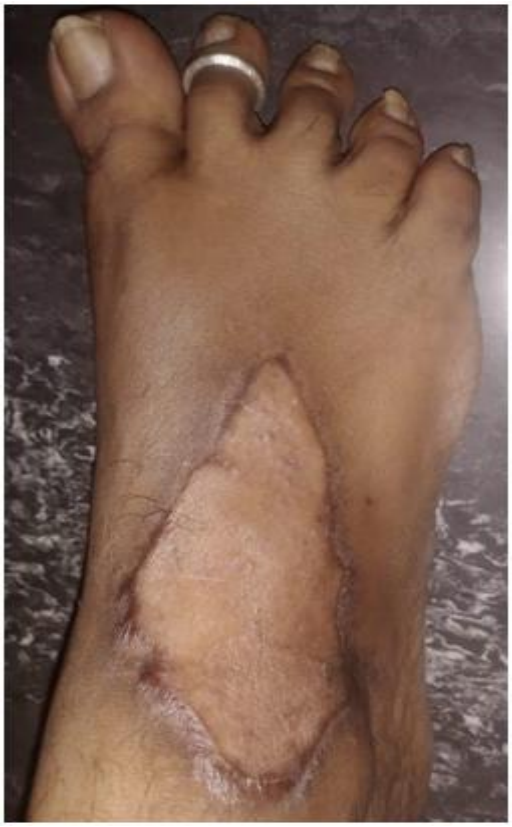

Figure 3: 16 Weeks Post Grafting

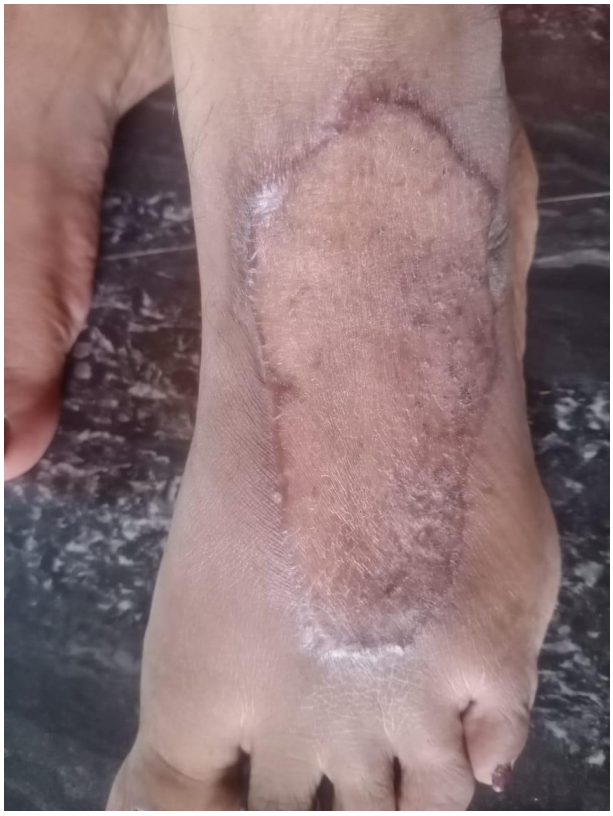

Figure 4: 24 Weeks Post Grafting

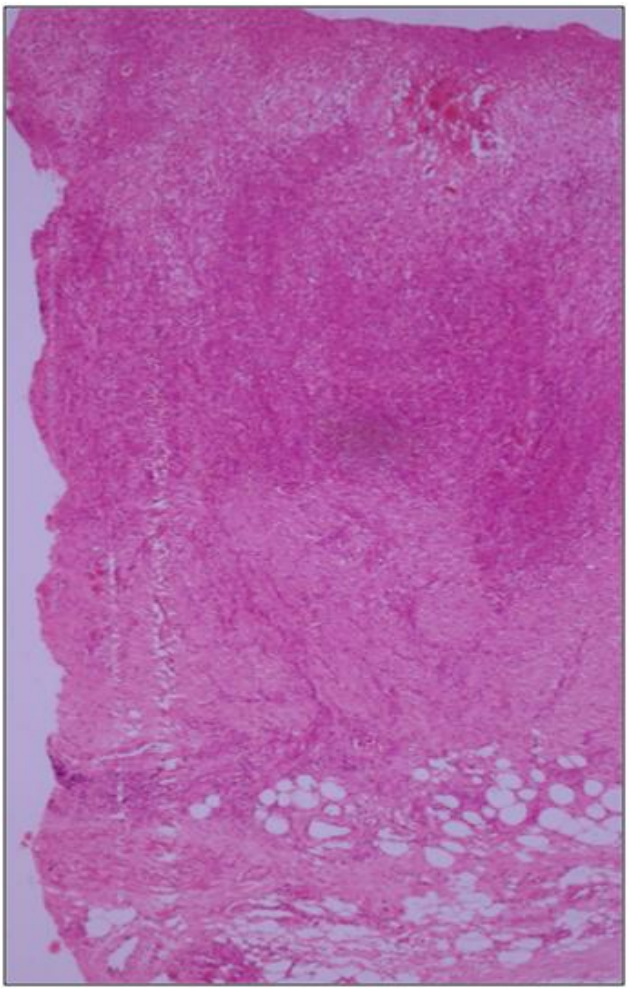

Figure 5: HISTOPATHOLOGY 100X VIEW-dense dermo-epidermal neutrophilic infiltrate and perivascular lymphocytic infiltrate