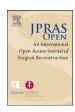


Contents lists available at ScienceDirect

JPRAS Open

journal homepage: www.elsevier.com/locate/jpra



Case Report

Perforator propeller flap for coverage of Achilles region defects caused by pyoderma gangrenosum

Chun Wa Fong*, Manuel Bento, Feng Jun Fang, Fong Kuong Pang, Io Hang Lio, Sut Sin Tong, Chou Kuan Hao

Department of Plastic and Reconstructive Surgery, Centro Hospitalar Conde São Januário, Estrada do Visconde de S. Januário, Macau SAR

ARTICLE INFO

Article history: Received 14 December 2021 Accepted 27 June 2022 Available online 6 July 2022

Keywords: Pyoderma gangrenosum Surgical treatment Propeller flaps

ABSTRACT

Pyoderma gangrenosum (PG) is a rare neutrophilic inflammatory skin disease. Systemic corticosteroid and immunosuppressive agents are the mainstay treatment. PG usually precludes a surgical approach due to pathergy phenomenon. Recent literatures show skin grafting and negative pressure wound therapy are safe if performed under adequate immunosuppression. We present a case of a 61-year-old male patient suffered from PG induced left posterior leg wound with Achilles tendon exposure. We made timely diagnosis and treated him with adequate immunosuppression therapy followed by perforator propeller flap for wound coverage. This case report emphasizes the need for high index of suspicion for PG diagnosis. Furthermore, with adequate immunosuppression, operative intervention may not be an absolute contraindication for PG.

© 2022 The Author(s). Published by Elsevier Ltd on behalf of British Association of Plastic, Reconstructive and Aesthetic Surgeons.

This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/)

E-mail address: jeffery.fong128@gmail.com (C.W. Fong).

^{*} Corresponding author.

Introduction

Pyoderma gangrenosum (PG) is a rare non-infectious, reactive neutrophilic inflammatory dermatosis with an estimated incidence of 3 to 10 cases per million people per year. It was first described in 1908 but its etiology is still not thoroughly understood. About one third of the cases are related to pathergy (an exaggerated skin injury occurring after minor trauma). The increased activity of polymorphonuclear leukocytes has been considered as the underlying pathophysiologic change of pathergy. PG clinical presentations are various and there are multiple subtypes, such as ulcerative, bullous, pustular, vegetative and post-surgical PG. It is a diagnosis of exclusion and immunotherapy is the mainstay treatment. Surgical intervention for PG is controversial because of pathergy reaction.

Herein, we report a case of a 61-year-old male who suffered from Achilles tendon-exposed wound caused by PG. We successfully reconstructed the wound with perforator propeller flap with adequate immunosuppression therapy.

Case presentation

A 61-year-old male presented with swelling, erythema, warm and pain on left posterior lower leg for two days. He punctured the painful area then had intermittent high-grade fever and antibiotics were initiated in private clinic for three days. The patient had no relevant medical or personal history.

At our emergency room, his left lower leg presented erythema, tenderness, with bloody and purulent exudate. Blood test revealed neutrophilic leukocytosis, high C-reactive protein and slightly increased procalcitonin. Incision drainage and debridement were done, Meropenem and Clindamycin were started. Vancomycin was added afterwards because wound culture showed Methicillin-resistant Staphylococcus aureus. After five-day course of Vancomycin, the ulcer continued deteriorating with severe pain, Achilles tendon exposure and wound size expanding to 12cmx8cm. The purulent necrotic ulcer was seen with a grayish, ragged and violaceous border, worsening with every minor debridement (Figs. 1 and 2). Pathology of the wound edge showed neutrophilic exudates. Negative swab culture was obtained. Due to the rapid progression of the painful necrolytic cutaneous ulcer with irregular violaceous undermined border, history of pathergy, histopathological findings, and other causes of ulceration excluded, we considered PG according to the diagnostic criteria.³ Empirical treatment with 40 mg prednisolone orally per day was initiated. Two days later, wound pain significantly improved and the ulcer was under control with less erythematous border. On the fifth day of steroid, we performed wound debridement, Integra® placement, and negative pressure wound therapy with instillation. Prednisolone dosage was subsequently tapered down the following two-week period to 10 mg per day. The patient then underwent a second operation for reconstruction. The devitalized part of Integra which lied over the tendon was removed. The left posterior tibial artery perforatorbased propeller flap was elevated and rotated 180° into the posterior lower leg defect. The donor-site defect was covered by split thickness skin graft.

Post-operatively, Cyclosporine 150 mg per day was initiated and we kept tapering and discontinued prednisolone in one week. The patient's leg wound healed after one month. He kept follow up in outpatient clinic and there were no signs of relapse (Fig. 3). We successfully covered the PG-caused Achilles tendon exposed wound without significant morbidity and deformity.

Discussion

Pyoderma gangrenosum (PG) is a rare neutrophilic inflammatory skin disease which remains a diagnosis of exclusion because of its non-specific clinical, laboratory and histopathological findings.⁴ Therefore, early diagnosis of PG is challenging and depends on high alert and recognition about the disease. This patient presented with infection signs at left lower leg first but the clinical condition did not respond well to empirical treatment and kept deteriorating. According to the Delphi diagnostic criteria for PG (Table 1), we reached the diagnosis for our patient in two weeks since his initial presentation.³ The Delphi diagnostic criteria was introduced in 2018 and consists of 1 major criterion and 8 minor criteria. The sensitivity and specificity for PG diagnosis are high when fulfilling 1 major criterion and 4 of 8 minor criteria.



Fig. 1. Patient's left posterior lower leg wound initially.

Treatment options for PG include topical and intralesional therapy, systemic corticosteroid and biologic agents.⁴ Systemic corticosteroid is considered to be the first-line treatment for most PG patients. Nevertheless, the PG wound healing process is long and less than half of the wounds healed after six-month immunosuppression treatment.⁵ Surgical intervention following adequate immunosuppressive therapy may accelerate healing time but clinical evidence is limited which only focus on split thickness skin grafting with negative pressure wound therapy and the timing of surgery varies.⁶⁻⁸ However, for the exposed Achilles tendon PG wound we described, prolonged tendon exposure would cause significant morbidity and skin graft is destined to fail to revascularize. Therefore, soon after



Fig. 2. The lower leg wound got worse rapidly with Achilles tendon exposure after incision drainage, debridement and antibiotics.

Table 1
Diagnostic criteria for classic ulcerative pyoderma gangrenosum.

Major criteria: Biopsy of ulcer edge demonstrating neutrophilic infiltrate

Minor criteria

- 1. Exclusion of infection
- 2. Pathergy
- 3. History of inflammatory bowel disease or inflammatory arthritis
- 4. History of papule, pustule, or vesicle that rapidly ulcerated
- 5. Peripheral erythema, undermining border, and tenderness at ulceration site
- 6. Multiple ulcerations, at least 1 on an anterior lower leg
- 7. Cribriform or "wrinkled paper" scar(s) at healed ulcer sites
- 8. Decreased ulcer size within 1 month of initiating immunosuppressive medication

clinical improvement with steroid initiation, we used acellular dermal matrix and negative pressure wound therapy with instillation to cover the wound temporally. Dermal matrix served as a transitional method before final reconstruction, allowing further medical control for PG and protecting the exposed tendon from desiccation.⁹

With immunosuppressive medication, the disease became quiescent and reconstruction surgery was planned. We chose perforator propeller flap for coverage of the wound. The term "propeller flap" was first introduced by Hyakusoku et al. in 1991 and was formally defined as an "island flap that reaches the recipient site through an axial rotation" during the Tokyo Consensus in 2011.^{10,11} It has been widely used in reconstruction of soft tissue defects in the middle and distal lower extremities, based on different main vessels, such as anterior tibial, posterior tibial, peroneal, lateral malleolar. Its advantages include minimizing the donor site morbidity, replacing like-with-like tissue and easier to perform without the need of microvascular anastomoses.



Fig. 3. Nine-month postoperative result.

Conclusion

Our case demonstrated that PG can be complicated with infection and exposure of underneath important structures. Consideration of a diagnosis of PG is indicated if the disease fails to respond as expected to therapy. With adequate immunosuppression, surgical intervention may not be an absolute contraindication for PG and perforator propeller flap is a versatile option in reconstruction of soft tissue defect over Achilles tendon region, replacing like-with-like and no need of microvascular anastomoses.

Funding

None declared.

Ethical approval

Not required.

Informed consent

Full written informed consent was sought for the publication of this case report and the associated clinical images.

Conflict of interest

None declared.

References

- Tolkachjov SN, et al. Postoperative pyoderma gangrenosum: a clinical review of published cases. Mayo Clin Proc. 2016;91(9):1267–1279.
- Braswell SF, Kostopoulos TC, Ortega-Loayza AG. Pathophysiology of pyoderma gangrenosum (PG): an updated review. J Am Acad Dermatol. 2015;73(4):691–698.
- 3. Emanual M, Chelsea M, et al. Diagnostic Criteria of Ulcerative Pyoderma Gangrenosum: a Delphi Consensus of International Experts. *JAMA Dermatol.* 2018 Apr 1;154(4):461–466.
- 4. Goldust M, et al. Diagnosis and novel clinical treatment strategies for pyoderma gangrenosum. Expert Rev Clin Pharmacol. 2020 Feb;13(2):157–161.
- Ormerod AD, et al. Comparison of the two most commonly used treatments for pyoderma gangrenosum: results of the STOP GAP randomised controlled trial. BMJ. 2015 Jun 12;350:h2958.
- 6. Pichler M, et al. 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):760–765.
- 7. Pichler M, et al. Systematic review of surgical treatment of pyoderma gangrenosum with negative pressure wound therapy or skin grafting. J Eur Acad Dermatol Venereol. 2017 Feb;31(2):e61–e67.
- 8. Eisendle K, et al. 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 [ul;9(7):405–425.
- Goshtasby PH, et al. A novel approach to the management of pyoderma gangrenosum complicating reduction mammaplasty. Aesthet Surg J. 2010 Mar; 30(2):186–193.
- 10. Hyakusoku H, Yamamoto T, Fumiiri M. The propeller flap method. Br | Plast Surg. 1991 |an;44(1):53-54.
- 11. Pignatti M, Ogawa R, et al. The "Tokyo" consensus on propeller flaps. Plast Reconstr Surg. 2011 Feb;127(2):716-722.