CASE REPORT

# Pyoderma Gangrenosum of the Hand: Unique Experience and Literature Review

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**Background:** Pyoderma gangrenosum (PG) is a reactive, noninfectious, neutrophilic dermatosis. Diagnosis of PG is based on exclusion, due to lack of availability of a confirmatory test. PG is not caused by infection or gangrene. Misdiagnosis or delayed diagnosis of PG can lead to devastating results.

**Case Presentation:** In this report, we present a patient with a delayed diagnosis of PG lesion on right hand. Despite initial surgical treatment, the wound was aggravated, and amputation was considered; however, it was eventually treated successfully with an autologous split thickness skin graft.

**Conclusions:** Knowledge of the PG is essential to actively consider PG in early stage to help facilitate immediate treatment and avoid unnecessary interventions that may worsen the outcome.

**Key words:** Hand; Infection; Pyoderma gangrenosum; Skin graft

## Introduction

Pyoderma gangrenosum (PG) is a reactive, noninfectious, neutrophilic dermatosis of unknown origin characterized by a spectrum of clinical presentations with variable courses.[1] PG is a diagnostic and therapeutic challenge. Because of the lack of the availability of confirmatory test, currently the diagnosis is made by exclusion of other similar ulcerative conditions.[2] At a rate of ~10%, misdiagnosis of pyoderma gangrenosum is not uncommon.[3] PG is known to often occur in association with a systemic disease, such as inflammatory bowel disease, metabolic syndrome, rheumatologic or hematological disorders, or even malignancies.[4] Recently, there have been reports of PG occurring after COVID-19 infection or vaccination.[5,6] We report a case of delayed diagnosis of PG of hand, without systemic disease, with frustrating soft tissue condition, and close to amputation. However, we managed to successfully treat this case with split-thickness skin graft.

## Case Report

A 79-year-old male presented with right hand index finger skin injury caused by sawing ~3 months ago. The patient had no systemic disease, as confirmed by an internal medicine physician and a bone marrow biopsy. However, he was vaccinated with the second dose of BNT162b2 COVID-19 vaccine ~6 months ago. He complained about worsening index finger pain and a thumb skin defect that occurred spontaneously without direct trauma ~1 week ago (Figure 1). On physical examination, severe pain and tenderness was observed around the wound and radiography of the hand showed no remarkable finding apart from the swelling of soft tissue. On laboratory examination, white blood cells (WBC) count was reported to be 11,450/ml (segmented neutrophils

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80.1%), erythrocyte sedimentation rate (ESR) was 100 mm/h, and C-reactive protein (CRP) value was 10.02 mg/dl. We initially diagnosed the patient with a skin defect with superimposed cellulitis and the initial treatment plan included intravenous antibiotics administration and wound care. Because of sustained fever and worsening symptoms, magnetic resonance imaging (MRI) with enhancement was performed; however, no remarkable abscess was found. MRI showed diffuse edema with mild enhancement of skin and subcutaneous fat layer, suggesting cellulitis. Debridement of devitalized tissue was performed in the ward until the sixth hospital day (HD). However, the wound spread rapidly through the entire hand and the wrist despite daily debridement, with swelling observed in the wound. Even in areas of tissue without debridement, hemorrhagic bullae were observed and appeared to be spreading. Moreover, the lesion disseminated to the entire body, including both legs (Figure 2). The various bacterial cultures performed with the sample from the wound were found out to be negative. However, rapid progression, including worsening pain, elevation of inflammatory index of laboratory finding, sustained fever, and disseminated wound suggested uncontrolled infection and sepsis. Therefore, we decided to perform incision of the wound to release the tissue pressure in order to decrease the pain and

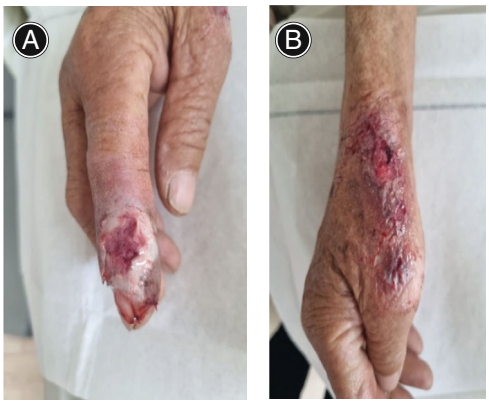


**FIGURE 1** (A) Initial wound in the index finger from sawing. (B) Spontaneous lesion of the thumb

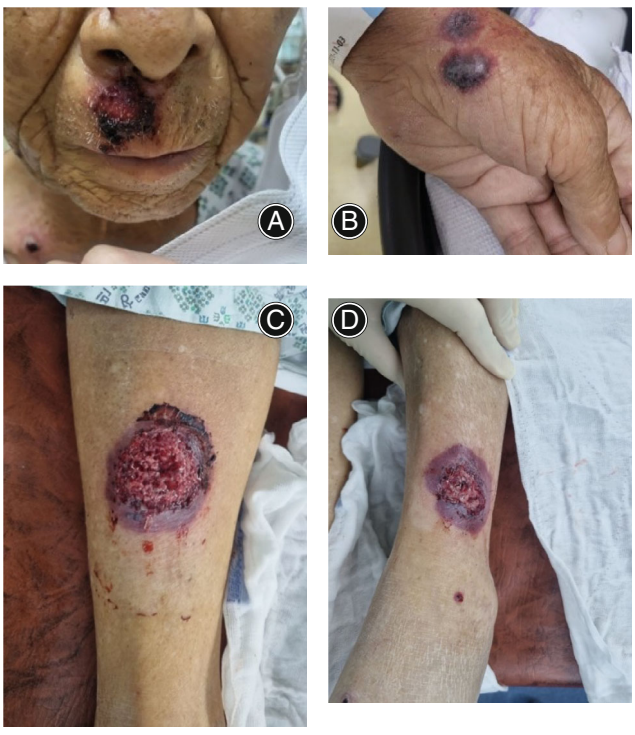


**FIGURE 2** Disseminated lesions. (A) Philtrum. Disseminated lesions. (B) Contralateral hand. Disseminated lesions. (C, D) Both leg lesions, anterior leg lesion is a feature of pyoderma gangrenosum (PG)

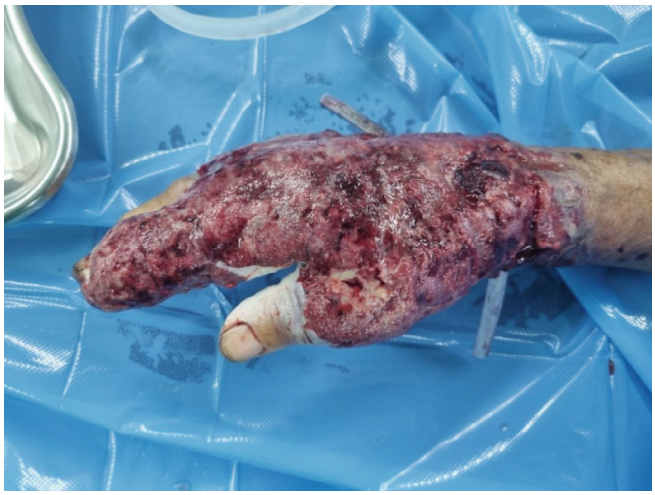

**FIGURE 3** After surgical incision and drainage, frustrating worsened wound shows pathergy pattern of pyoderma gangrenosum

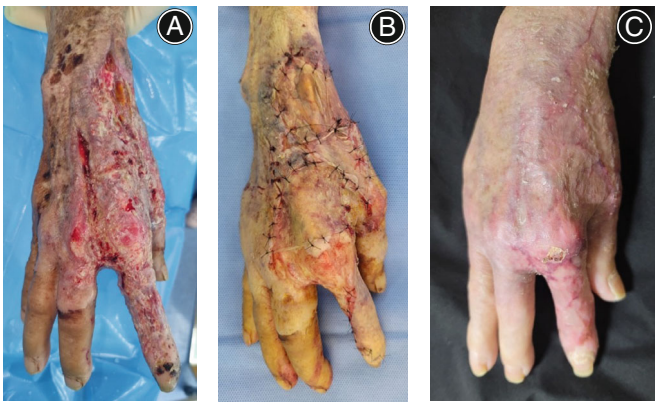


**FIGURE 4** (A) Bone and tendon were not exposed in the photograph at 2 weeks after the modified treatment. (B) Split-thickness skin graft (STSG) was performed. (C) 2 months after STSG

drain the abscess. During the surgery, histologic examination and bacterial culture were requested. Operative findings indicated that both flexor and extensor tendons were intact; however, the subcutaneous layer oozed pus that was sent for pathological examination. Based on the clinical findings, we suspected the diagnosis to be pyoderma gangrenosum (PG). Because infection and pyoderma gangrenosum are treated differently, no further incision or debridement other than irrigation cleansing after surgery was performed until pathologic findings were confirmed. Worsening wound condition after incision and drainage showed pathergy character of PG (Figure 3). The pathologist confirmed abscess and inflammatory change of tissue harvested from the center of the wound, and marginal tissue findings revealed neutrophilic aggregation with necrosis in the upper dermis and subcutis. These finding supported the diagnosis of PG. For treatment of PG, systemic steroid, cyclosporine, and local steroid ointment were used. The swelling and pain decreased dramatically after cleansing with normal saline and local steroid ointment was applied without debridement. There was sufficient soft tissue coverage of bone and tendons. However, the skin defect on the surface was too wide; therefore, split-thickness skin graft was needed to cover the wound. Because of pathergy feature of PG, we were concerned about skin graft survival, but decided to use autologous skin donor from the thigh. Though the graft survived, owing to prolonged immobilization, the range of motion in the finger decreased (Figure 4). After 6 months of rehabilitation, the patient was able to return to activities of daily living (Figure 5).

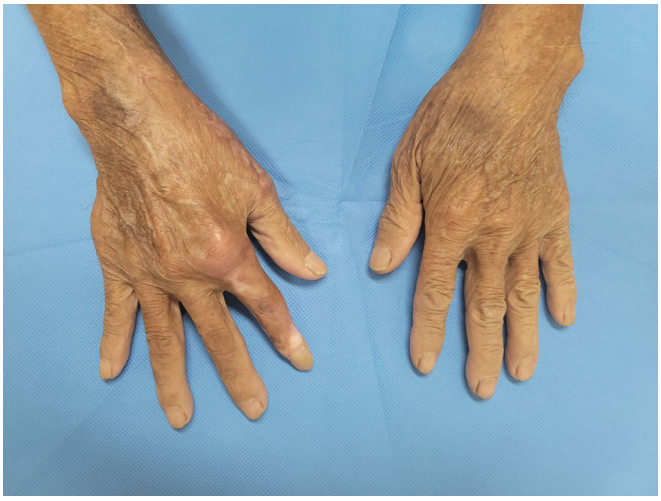

**Figure 5** After 6 months of rehabilitation, the patient was able to return to activities of daily living

## Discussion

Amisdiagnosis or delayed diagnosis of pyoderma gangrenosum (PG) can lead to increased morbidity and death because of its pathergy character. There are several reports where amputation of the affected part had to be performed.[2] In this case, we had difficulty in the diagnosis because of the rarity of PG, its multiple variants and their own pattern, and a diagnosis based on exclusion.

Although there is a diagnostic tool proposed by Su *et al*., there is still the need for exclusion of other ulcerations.[7] Maverakis *et al*. proposed new diagnostic criteria for ulcerative PG that required only the exclusion of infection and shows sensitivity and specificity of 86% and 90%, respectively.[8] Based on these new diagnostic criteria, a diagnosis of PG is no longer a diagnosis of exclusion; one major criterion requires biopsy of ulcer edge demonstrating a neutrophilic infiltrate, and four of eight minor criteria including exclusion of infection, three of patient history, three of photographic evidence, response to immunosuppressive medication. Various PG variants are known: ulcerative, bullous, pustular, vegetative, peristomal, post-surgical. Each subtype has a common location and associated disease.[9] There was a linear dead soft tissue injury from scalpel incision during surgery; otherwise, the untouched soft tissue was viable. Caution must be exercised in determining the treatment methods when the diagnosis is uncertain. In up to 50% of cases, PG is known to be associated with systemic disease, including malignancy. Thus, clinicians should try to ascertain the possible underlying disease. Conversely, the diagnosis of a systemic disease in patients can help in their PG diagnosis. For treatment, the pathophysiologic mechanisms have not yet been clearly elucidated. It relies on poorly evidenced publications, such as case series. Although there is insufficient evidence for the ideal medical therapy and duration of treatment, management is based on clinical severity and extent of the PG. Topical

**TABLE 1 Reports published in the literature on the surgical treatment of pyoderma gangrenosum in the hand using skin graft**

| No | Author | Patients' age/sex | Location | Surgical treatment | Systemic treatment |
|---|---|---|---|---|---|
| 1 | Gérard *et al*. (1988) | M/41 | Left hand | STSG (Splint thickness skin grafting) | CS (Cortical steroid), Plama exchange |
| 2 | Mowlds *et al*. (2013) | F/61 | Both hand | STSG and NPWT (Negative pressure wound therapy) | Acetic acid dressing and HBO (Hyperbaric oxygen) |
| 3 | Leitsch *et al*. (2016) | M/54 | Hand and forearm | STSG and NPWT | CS |
| 4 | Hradil *et al*. (2017) | M/72 | Right hand and forearm | STSG | CS |

corticosteroids are often tried initially at localized PG, but in more severe cases, systemic treatment, using systemic corticosteroids and immunosuppressive medications, including cyclosporin or biologic therapies targeting a number of cytokines, are used.[1,9] Pathergy, an exaggeration of soft tissue injury occurring after minor trauma, poses another difficulty in the treatment of soft tissue coverage such as flap or skin graft. However, there are only four reports published in the literature on the surgical treatment of PG in the hand using skin graft (Table 1). There is no consensus for perioperative medical regimen to decrease donor site morbidity, and the recurrence rate has been reported to be 16.7%.[10]

### *Conclusion*

Pyoderma gangrenosum (PG) is usually a rare disease. However, knowledge of the disease is essential to actively consider PG in early stage to help facilitate immediate treatment and avoid unnecessary interventions that may worsen the outcome.

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## References

**1.** George C, Deroide F, Rustin M. Pyoderma gangrenosum - a guide to diagnosis and management. Clin Med (Lond). 2019;19:224–8. https://doi.org/10.7861/clinmedicine.19-3-224

**2.** Haag CK, Nutan F, Cyrus JW, Satpathy J, Shinkai K, Ortega Loayza AG. Pyoderma gangrenosum misdiagnosis resulting in amputation: a review. J Trauma Acute Care Surg. 2019;86:307–13. https://doi.org/10.1097/TA.0000000000002096

**3.** Weenig RH, Davis MD, Dahl PR, et al. Skin ulcers misdiagnosed as pyoderma gangrenosum. N Engl J Med. 2002;347:1412–8. https://doi.org/10.1056/NEJMoa013383

**4.** Bennett ML, Jackson JM, Jorizzo JL, Fleischer AB Jr, White WL, Callen JP. Pyoderma gangrenosum. A comparison of typical and atypical forms with an emphasis on time to remission. Case review of 86 patients from 2 institutions. Medicine (Baltimore). 2000;79:37–46. https://doi.org/10.1097/00005792-200001000-00004

**5.** Barry M, AlRajhi A, Aljerian K. Pyoderma Gangrenosum induced by BNT162b2 COVID-19 vaccine in a healthy adult. Vaccines (Basel). 2022;10:20220107. https://doi.org/10.3390/vaccines10010087

**6.** Syed K, Chaudhary H, Balu B. Pyoderma gangrenosum following COVID-19 infection. J Community Hosp Intern Med Perspect. 2021;11:601–3. https://doi.org/10.1080/20009666.2021.1958492

**7.** Su WP, Davis MD, Weenig RH, et al. Pyoderma gangrenosum: clinicopathologic correlation and proposed diagnostic criteria. Int J Dermatol. 2004;43:790–800. https://doi.org/10.1111/j.1365-4632.2004.02128.x

**8.** Maverakis E, Ma C, Shinkai K, Fiorentino D, Callen JP, Wollina U, et al. Diagnostic criteria of ulcerative pyoderma Gangrenosum: a Delphi consensus of international experts. JAMA Dermatol. 2018;154:461–6. https://doi.org/10.1001/jamadermatol.2017.5980

**9.** Alavi A, French LE, Davis MD, Brassard A, Kirsner RS. Pyoderma Gangrenosum: an update on pathophysiology, diagnosis and treatment. Am J Clin Dermatol. 2017;18:355–72. https://doi.org/10.1007/s40257-017-0251-7

**10.** Haag CK, Bacik L, Latour E, Morse DC, Fett NM, Ortega-Loayza AG. Perioperative management of pyoderma gangrenosum. J Am Acad Dermatol. 2020;83:369–74. https://doi.org/10.1016/j.jaad.2020.01.002