



Inflammation and infection

Broadening the differential: A case of penile pyoderma gangrenosum

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ABSTRACT

Pyoderma gangrenosum can be a challenging diagnosis for even the most experienced clinician. Misdiagnosis can lead to delays in appropriate treatment and unwarranted debridement that can increase the severity of the disease. Penile pyoderma gangrenosum (PG) is a rare presentation of this pathologic process. We describe the diagnostic workup and successful treatment of advanced penile PG in a 42-year-old male with a history of penile fracture who presented with delayed wound healing and multiple unsuccessful urologic surgeries. This case demonstrates the importance of keeping a broad differential, including PG, in order to avoid delays to appropriate care.

Introduction

Pyoderma gangrenosum (PG) is a noninfectious inflammatory condition characterized by neutrophilic dermatosis.¹ PG most commonly affects the lower extremities, but has also been seen on the face, neck, and rarely in penile skin.¹ PG is a diagnosis of exclusion and can be delayed due to misdiagnosis of malignancy or a sexually transmitted infection that fails to respond to standard therapy.² It is, however, important to keep PG in the differential diagnosis as serious complications, such as urethral fistulas, have occurred when PG is not diagnosed and managed in a timely manner.¹ Treatment of PG is driven by immunosuppressive agents including cyclosporine and corticosteroids.³ Although PG resolves with appropriate treatment, the rarity and nonspecific presentation of penile PG can lead to mismanagement and extension of the lesion.² In this case report we document appropriate diagnostic work up and successful treatment of advanced penile PG with immunomodulators including prednisolone, adalimumab and cyclosporine.

Case presentation

A 42-year-old, male with a history of type II diabetes mellitus (Hgb A1c 7.4–8.6) and a 20 pack year smoking history and buccal mucosal urethroplasty two years earlier re-presented in 2014 for a bulge on the left side of his penis with erection, associated with left curvature and pain. He first noticed the bulge 4 months prior to presentation after hearing a crack followed by pain and swelling during traumatic

intercourse. Imaging studies confirmed a defect in the left tunica without urethral injury. He underwent attempted re-approximation of the urethral edges and primary penile fracture repair. Intraoperative observation during repair demonstrated severe corporal fibrosis and scarring that was successfully reapproximated and closed in a standard two layer fashion.

Over the next 3 years, he continued to have localized erythema edema, and drainage from the fracture site. The decision was made to bring him to the operating room to attempt secondary repair and repeat skin closure. Intraoperative biopsy of the site showed benign granulation tissue. Workup for causes of being immunocompromised, including, IgG, IgM, hepatitis, and HIV panels, were all normal. Six months after his secondary repair the wound had not healed and the previously noted erythema, edema, and ulceration had spread to the peno-scrotal junction. He was referred for localized wound care and hyperbaric O₂ therapy. Wound cultures collected before starting treatment were notable for *Pseudomonas aeruginosa* and the patient was initiated on a 2 week course of oral ciprofloxacin. The degree of skin ulceration continued to worsen with expansion from initial size of 2–3 cm to a larger 5–6 cm ulcer and the decision was made to refer for a secondary opinion in urology.

Examination at this time demonstrated a well-defined ulceration to the level of Buck's fascia over the left penis and penoscrotal border accompanied by fibrinous exudate and beefy erythematous wound edges that were extremely tender to palpation (Figs. 1–2).

Given the worsening wound ulceration and otherwise negative workup, PG became a consideration and the patient was appropriately

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Fig. 1. Dorsal penile wound at initial referral for second opinion.



Fig. 2. Lateral penile wound at initial referral for second opinion.

referred for dermatological evaluation. He was started on a 3 week taper of prednisone 20mg and adalimumab 40mg/0.8 mL every 14 days with gradual improvements over the next 2–6 weeks. Eight weeks after initiating treatment the lesion was deemed sufficiently healed and oral prednisone was discontinued in favor of a prolonged course of cyclosporine 100mg 3 times a day for 1 month.

Twelve weeks after initiation of immunomodulatory therapy the lesion appeared fully resolved with return of well vascularized fibrotic tissue (Fig. 3). Residual leftward angulation of the penis and healed wound bed can be seen in the picture below. The patient reported zero erectile function and SHIM score of zero at this point, but did report maintained flexibility of the right corporal body with bending of the penis. He planned to purchase a VED to attempt aggressive penile rehabilitation, trial PDE5 inhibitors, and continue to follow up for treatment of his erectile dysfunction.

Discussion

Pyoderma gangrenosum is a neutrophil associated ulcerative skin disease. Patients develop painful ulcerative skin lesions with histopathologic evidence of neutrophilic invasion in the absence of other infectious causes.⁴ One of the greatest challenges to diagnosis and management of PG is that it is considered a diagnosis of exclusion. Without laboratory test with which it can be reliably identified, clinicians must rely on a thorough history and physical exam, appropriate microscopic and histopathological testing, and diagnostic imaging to rule out other systemic diseases that can present similarly or alongside PG.

PG of penile skin is rare, but can be especially difficult to identify because PG can easily be confused with a number of STIs that present with penile ulceration. Therefore, prompt evaluation for STIs should be done at initial presentation of PG. It is especially important to inquire about previous urologic surgeries, as studies have cited an association between local trauma, cancer treatment, and surgery as playing a role in the development of PG.¹ Furthermore, extensive debridement of PG ulcers can paradoxically worsen the lesion in a process termed pathergy.

Management of this condition can be a great challenge, as no particular immune modulating regimen has been identified as standard of care.⁴ Instead treatment decisions are determined on a case by case basis. In many cases, topical therapy with medications like imiquimod are sufficient to allow for healing of ulceration if limited to a small unifocal area.¹ Advanced cases, on the other hand, are more likely to require systemic therapy with corticosteroids, calcineurin inhibitors, or monoclonal antibodies inactivating TNF-alpha. It is especially important for patients who develop PG to be followed long term to ensure no further recurrences develop.²

Conclusion

This case helps to illustrate the need to consider PG in the case of chronic ulcerating, non-healing wounds of the penis. This patient underwent multiple procedures for delayed wound healing, which may



Fig. 3. Recovered dorsal and lateral penile wounds 12 weeks post initiating immunomodulatory therapy.

have paradoxically accelerated the severity of his disease as well as

exposed him to unnecessary risks of surgery. Earlier diagnosis of PG could have avoided extra surgeries for the patient. Ultimately the patient has successfully healed his wounds and recovered from the PG ulcerations once appropriate treatment was initiated.

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