Pyoderma gangrenosum: An uncommon cause of nonsexually acquired genital ulcer disease

Taru Garg, Himadri Himadri, Amit Kumar Meena, Vibhu Mendiratta, Shilpi Agarwal[1]
Departments of Dermatology and STD and [1]Pathology, Lady Hardinge Medical College, New Delhi, India

Address for correspondence:
Dr. Himadri Himadri, Department of Dermatology and STD, Lady Hardinge Medical College, New Delhi - 110 001, India.
E-mail: himadri2sinha@yahoo.com

Abstract

Pyoderma gangrenosum (PG) is a rare neutrophilic dermatosis affecting various sites, isolated genital PG being an uncommon presentation. We report a case of a 50-year-old diabetic male who presented with 2 penile ulcers. Extensive evaluation was done for sexually and nonsexually transmitted infections, malignancy, drug-induced vasculitis, and immunobullous etiology. A diagnosis of PG was made based on the clinical findings and histopathological exclusion of other causes. The patient showed a rapid response to prednisolone, dapsone, and colchicine. This report highlights the importance of keeping PG as a differential diagnosis in cases of genital ulcers which may mimic other sexually transmitted infections.

Key words: Nonsexually acquired genital ulcer, penile ulcers, pyoderma gangrenosum

Introduction

Pyoderma gangrenosum (PG) is a rare neutrophilic dermatosis (incidence: 0.73/1,00,000 person years) affecting various sites, isolated genital PG being an uncommon presentation. Almost 75% cases are associated with underlying systemic diseases such as inflammatory bowel disease, arthritis, and hematological disorders.[1] We hereby report a case of isolated penile PG without systemic involvement.

Case Report

A 50-year-old gentleman presented with 2 ulcers over the glans penis for 10 days. They started as minimally painful papules which rapidly ulcerated with increase in size. There was no history of discharge, bleeding, fluid-filled lesions, trauma, recurrent oral ulcers, redness of eyes, joint pains, gastrointestinal symptoms, urethral discharge, fever, loss of weight, or appetite. The patient was married and gave a history of multiple extramarital contacts with commercial sex workers, last contact being 15 days before the onset of the ulcers. There was no history of sexually transmitted disease (STD) in partner. He was a known diabetic on irregular treatment for 5 years.

General examination was unremarkable. Peripheral pulses were palpable. There was no lymphadenopathy. Glans penis had two well-defined tender, indurated ulcers with necrotic grayish-white slough, regular margins, undermined edges, erythematous border- measuring 3 cm × 2 cm and 0.5 cm × 0.5 cm [Figure 1a]. They did not bleed on touch. Rest of the mucosa, skin, palms, soles were normal.

Laboratory investigations

Gram's stain revealed multiple polymorphonuclear leukocytes, no organisms. Tzanck smear showed no multinucleated giant cells or acantholytic cells. Screening for HIV, Hepatitis B and C, syphilis (VDRL and TPHA), and herpes simplex virus serology were nonreactive. His fasting, postprandial sugars, and HbA1c were

Access this article online

Quick Response Code:

Website: www.ijstd.org

DOI: 10.4103/ijstd.ijstd_35_21

How to cite this article: Garg T, Himadri H, Meena AK, Mendiratta V, Agarwal S. Pyoderma gangrenosum: An uncommon cause of nonsexually acquired genital ulcer disease. Indian J Sex Transm Dis 2022;43:66-7.

Submitted: 14-Apr-2021 **Revised:** 18-Jul-2021
Accepted: 20-Sep-2021 **Published:** 07-Jun-2022

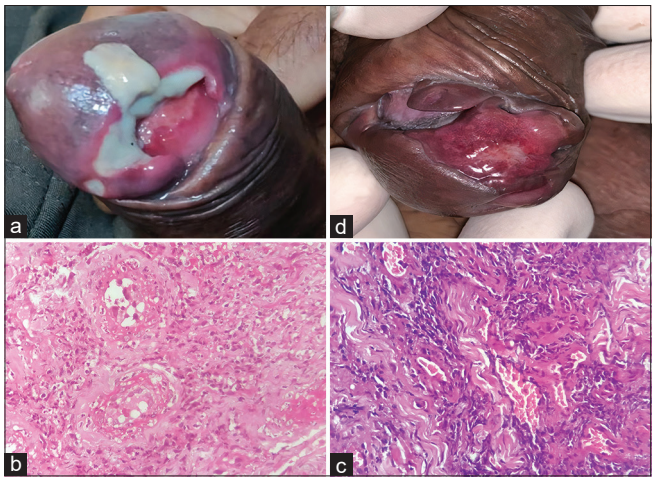

Figure 1: (a) Two ulcers with necrotic slough over the glans penis. (b) Photomicrograph of first biopsy showing acute inflammatory infiltrate and fibrinoid necrosis of vessels (×40). (c) Photomicrograph of repeat biopsy showing mixed inflammatory cell infiltrate (×40). (d) Healing ulcers showing healthy granulation tissue after treatment with immunomodulators

elevated (314 mg/dl, 362 mg/dl, and 7.3%, respectively). Pus culture for *Haemophilus ducreyi* was sterile. The Mantoux test was positive (15 mm × 17 mm). Skin biopsy [Figure 1b] showed acanthosis, fibrinoid necrosis in vessels, acute inflammatory cell infiltrate, and granulation tissue in the dermis along with necrotic debris. There was no evidence of dysplasia or malignancy. Repeat biopsy showed large areas of ulceration. Dermis showed granulation tissue with dense mixed inflammatory infiltrate of neutrophils, lymphocytes, and occasional plasma cells; there is no evidence of vasculitis, epithelioid cell granuloma, Crohn's disease, or malignancy [Figure 1c].

Ziehl–Neelsen staining did not show acid-fast bacilli, and culture for typical, atypical mycobacteria and tissue CBNAAT were negative. Chest X-ray was normal and contrast-enhanced computed tomography abdomen showed 16 mm × 10 mm right inguinal lymph node; no other significant abnormality. Pathergy test, stool for occult blood, and ANA were negative. Serum protein electrophoresis was normal.

With the initial acute presentation and history of high risk behavior, considering a differential diagnosis of primary chancre with mixed anerobic infection, chancroid, or herpes genitalis, we empirically treated him with local cleansing and oral antibiotics (metronidazole and ciprofloxacin). Metformin and teneligliptin were started. After ruling out STD, we considered differentials of noduloulcerative genital tuberculosis, atypical mycobacterial ulcer, malignancy, and PG. We diagnosed PG based on clinical findings and exclusion of other causes. We started him on dapsone 100 mg once daily and colchicine 0.5 mg thrice daily. After an initial response, the patient discontinued treatment by himself and developed another ulcer near the meatal opening. He was restarted on dapsone and colchicine. Prednisolone was added for 6 weeks with a maximum of 40 mg daily with which he showed rapid improvement [Figure 1d].

Discussion

PG is an uncommon neutrophilic disease, pathophysiology of which is incompletely understood. It involves a complex interplay of genetic influence, dysregulation in innate immunity, and neutrophil dysfunction.[2,3] It can affect all age groups with no sex predilection.[3] Clinical variants include classic ulcerative, bullous, pustular, vegetative, peristomal, etc. Various diagnostic criteria have been proposed although validated criteria are still lacking.[3] PG is an uncommon disease affecting all age groups. Diagnosis is made by excluding other causes. Pathergy may be seen in 20%–30% patients.[2] Management includes avoidance of triggers, local wound care, and immunomodulatory therapies.[3]

Few cases of isolated genital PG have been reported with only 4 from India.[4-7] Our patient responded well to a combination of therapy with dapsone, colchicine, and oral prednisolone in addition to local wound care. This case is presented to highlight the importance of keeping PG as a differential diagnosis of penile ulcers refractory to usual treatment modalities.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. Langan SM, Groves RW, Card TR, Gulliford MC. Incidence, mortality, and disease associations of pyoderma gangrenosum in the United Kingdom: A retrospective cohort study. J Invest Dermatol 2012;132:2166-70.
2. Braswell SF, Kostopoulos TC, Ortega-Loayza AG. Pathophysiology of pyoderma gangrenosum (PG): An updated review. J Am Acad Dermatol 2015;73:691-8.
3. 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.
4. Rathod SP, Padhiar BB, Karia UK, Shah BJ. Penile pyoderma gangrenosum successfully treated with topical Imiquimod. Indian J Sex Transm Dis AIDS 2011;32:114-7.
5. Philip M, Samson JF, Simi PS, Nandakumar G, Mathew B. Penile pyoderma gangrenosum. Indian J Sex Transm Dis AIDS 2013;34:138-40.
6. Bhat RM, Saldanha CS, Dandakeri S, Kambil SM. Genital pyoderma gangrenosum. Indian J Sex Transm Dis AIDS 2014;35:171-2.
7. Kambil SM, Bhat RM, Dandekeri S. Granulomatous pyoderma gangrenosum of the genitalia in the absence of Crohn's disease. Indian J Sex Transm Dis AIDS 2015;36:67-9.