Penile ulcerative pyoderma gangrenosum: A rare entity

Jalpa Kailashbhai Patel, Devna Subramonia Pillai, Priyanka Bodar, Pragya Ashok Nair
Department of Dermatology, Venereology and Leprosy, Pramukhswami Medical College, Shree Krishna Hospital, Bhaikaka University, Karamsad, Gujarat, India

Address for correspondence:
Dr. Pragya Ashok Nair, Department of Dermatology, Venereology and Leprosy, Pramukhswami Medical College, Shree Krishna Hospital, Bhaikaka University, Karamsad - 388 325, Gujarat, India.
E-mail: drpragash2000@yahoo.com

Abstract

Pyoderma gangrenosum (PG) is a rare, chronic, ulcerative, neutrophilic, and inflammatory skin disease. It most commonly affects the lower limb, may affect peristomal skin, and rarely involves mucosal and internal sites. Genital involvement has been rarely reported. Hereby, we report a case of penile PG in a 70-year-old male treated with oral steroids.

Key words: Neutrophilic dermatosis, penile, pyoderma gangrenosum

Introduction

Pyoderma gangrenosum (PG) is an ulcerating neutrophilic dermatosis associated with inflammatory bowel disease, hematologic disorders, and malignancy. Genital PG is a diagnosis of exclusion. It should be suspected in any patient having ulcerative penile lesions with negative diagnostic evaluation for infectious or other inflammatory conditions such as syphilis, herpes simplex, mycobacterial ulceration, donovanosis, squamous cell carcinoma (SCC), cutaneous Crohn's disease, drug reaction, Behcet's disease, and ulcerating sarcoidosis. It can also be suspected who fails to respond to adequate antibacterial or antiviral therapy.[1] A complete diagnostic workup to exclude all the above possible causes is a must to establish the early diagnosis to avoid unnecessary or potentially harmful interventions.

Case Report

A 70-year-old married male presented with painful genital ulcer for 20 days. Initially, the patient developed single skin-colored elevated lesion over the shaft of the penis which gradually ruptured leaving behind a raw area associated with clear fluid discharge. The patient was admitted to surgery ward for nonhealing ulcer, was given oral antibiotic and anti-inflammatory medications but did not improve so was referred to the dermatology outpatient department for further evaluation. Reference was done to the dermatology department. No history of any drug ingestion before development of ulcer was present. On history, he denied any extramarital contact. He was not a known diabetic or hypertensive, but his HBa1c was 6.7 on admission. Cutaneous examination showed single circular ulcer of 2 cm × 3 cm in size with undermined edges, granulomatous base, and serous discharge over the ventral aspect of the penis [Figure 1]. There was no inguinal or femoral lymphadenopathy. The ulcer did not bleed on touch. Giemsa stain did not reveal bacillary or coccobacillary Donovan bodies. Gram stain prepared from the ulcer was negative. Biopsy was taken keeping donovanosis, sarcoidosis, PG, cutaneous tuberculosis, and SCC penis as differential diagnosis. Histopathologic examination from the ulcer edge showed granulation tissue with moderate to intense mixed polymorphonuclear, lymphocytic and plasmacytic inflammatory infiltrate, many congested blood vessels, and vast areas of hemorrhage and necrosis [Figure 2a and b]. The routine laboratory investigation, rapid plasma reagin, X-ray chest PA view, enzyme-linked immunosorbent assay-HIV, HBsAg, anti-HCV, urine routine, and microscopy were negative. On the basis of clinical examination and other investigations which ruled out other conditions, an alternate diagnosis of PG was made. The patient was treated with oral prednisolone 20 mg once a day and then in tapering doses, for a month and topical soframycin cream. Lesions healed within 15 days [Figure 3].

Discussion

PG was described by Brunsting in the year 1930.[2] It is a rare, noninfectious, and idiopathic neutrophilic dermatosis commonly associated with underlying systemic disease most often affecting the lower extremities but may involve the face, neck, and rarely scrotum or penis as seen in the present case.[3]

Access this article online

Quick Response Code:

Website: www.ijstd.org

DOI: 10.4103/ijstd.ijstd_115_22

How to cite this article: Patel JK, Pillai DS, Bodar P, Nair PA. Penile ulcerative pyoderma gangrenosum: A rare entity. Indian J Sex Transm Dis 2023;44:77-9.

Submitted: 28-Nov-2022 **Revised:** 13-Jan-2023
Accepted: 19-Jan-2023 **Published:** 06-Jun-2023

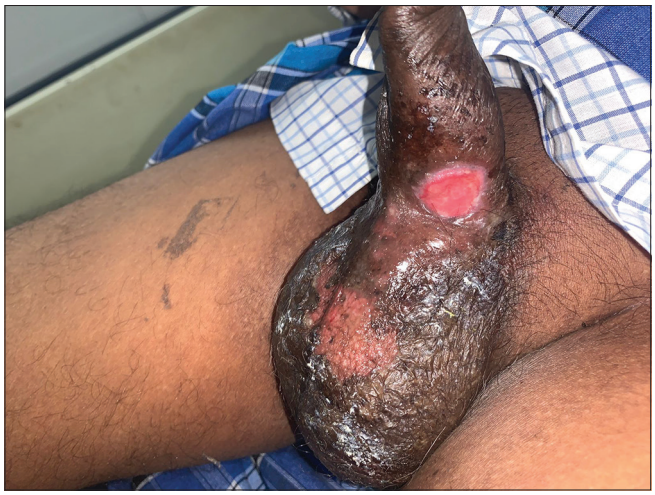

Figure 1: Single circular ulcer of 2 cm × 3 cm in size with undermined edges, granulomatous base, and serous discharge over the ventral aspect of the penis

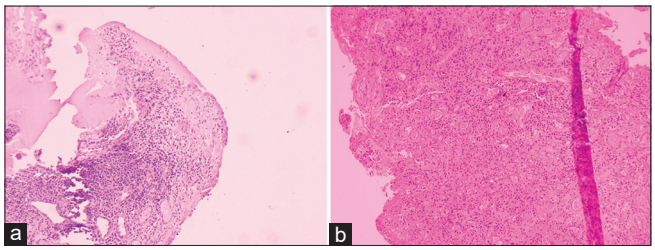

Figure 2: (a) Histopathology showed granulation tissue with moderate to intense mixed polymorphonuclear, lymphocytic and plasmacytic inflammatory infiltrate, many congested blood vessels, and vast areas of hemorrhage and necrosis (H and E stain × 10). (b) ×40

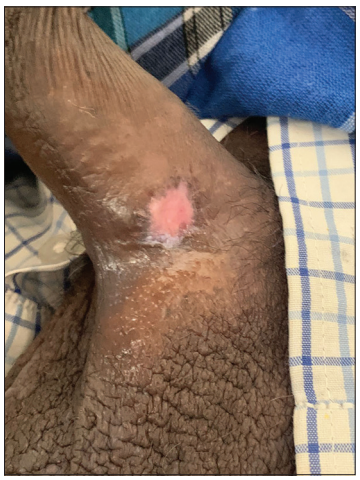

Figure 3: Healed ulcer after 15 days

Although it is idiopathic, in 25%–50% of patients, an underlying immunologic abnormality may exist.[4] The most common associations are inflammatory bowel disease (ulcerative colitis and Crohn's disease), arthritis (seronegative arthritis and rheumatoid arthritis), and hematologic diseases (myelogenous leukemia, hairy cell leukemia, myelofibrosis, and monoclonal gammopathy). It involves a complex interplay of genetic influence, dysregulation in innate immunity, and neutrophil dysfunction.[5] It can also be associated with other neutrophilic dermatoses such as Behçet's disease, subcorneal pustular dermatosis, and Sweet's syndrome. In our patient, no underlying systemic associations were found which is suggestive of the primary idiopathic form.

The extension of lesions in response to trauma or surgical debridement, termed pathergy, is a hallmark of PG.

Penile PG may occur following local trauma such as urological surgery, treatment for cancer, ulcerative colitis, or chronic lymphocytic leukemia. The disease begins with small tender papules or pustules that evolve into painful ulcers with characteristic violaceous undermined edges. Lesions may be solitary or multiple and heal with atrophic cribriform scar. It has a variety of clinical presentations such as ulcerative, pustular, bullous, and vegetative forms.[6]

Thus our case can be labeled as idiopathic ulcerative penile PG. It is important to rule out other causes of genital ulcer before reaching the rare diagnosis of PG. Painful genital ulcer of herpes genitalis start as grouped vesicles but rapidly become pustular and ulcerate. If secondary bacterial infection invades, the ulcer of donovanosis gets painful which is usually painless, beefy red in color and bleeds easily on touch. Tuberculous ulcer presents as papulonecrotic ulcer mainly on the buttocks, whereas sarcoidosis presents as chronic painful erythematous induration of the penis with several subcutaneous nodules and cutaneous ulcer. Other associated systemic symptoms are there with cutaneous tuberculosis and sarcoidosis. Penile SCC presents as a small area of induration and erythema or a large ulcerating and infiltrative lesion associated with foul odor and bleeding.

A report of a 70-year-old man presenting with indurated penile ulcers of 1-month duration showed features of PG in biopsy after inadequate response to doxycycline similar to our case.[7] Penile PG represents one of the classic ulcerative PG or superficial granulomatous pyoderma. Histopathological features although not specific in PG are important to rule out other causes of ulceration.

Oral corticosteroid therapy is the first-line treatment. Topical and intralesional corticosteroids or tacrolimus may be used in the superficial or pustular forms.[8,9] A case of a 24-year-old male patient living with HIV/AIDS (PLHIV) diagnosed as penile PG responded well with topical imiquimod 5% cream is reported. Other treatment options are topical imiquimod and cyclosporine.[1] Our patient showed excellent response to oral prednisolone and showed dramatic improvement within 15 days.

Conclusion

Although penile PG is rare and diagnosis of exclusion, one should keep in mind that any nonhealing painful penile lesion with negative laboratory and histopathological evaluations can be PG, as early recognition is critical to avoid unnecessary or potentially harmful interventions.

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. 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.
2. Ruocco E, Sangiuliano S, Gravina AG, Miranda A, Nicoletti G. Pyoderma gangrenosum: An updated review. J Eur Acad Dermatol Venereol 2009;23:1008-17.
3. Ecra E, Ahogo KC, Sangaré A, Kaloga M, Kassi K, Kouamé K, *et al*. An uncommon localization of pyoderma gangrenosum on the penis of an HIV infected patient in the Ivory Coast. Bull Soc Pathol Exot 2009;102:85-7.
4. Wollina U. Pyoderma gangrenosum – A review. Orphanet J Rare Dis 2007;2:19.
5. 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 AIDS 2022;43:66-7.
6. Hadi A, Lebwohl M. Clinical features of pyoderma gangrenosum and current diagnostic trends. J Am Acad Dermatol 2011;64:950-4.
7. Philip M, Samson JF, Simi PS, Nandakumar G, Mathew B. Penile pyoderma gangrenosum. Indian J Sex Transm Dis AIDS 2013;34:138-40.
8. Larsen CG, Thyssen JP. Pustular penile pyoderma gangrenosum successfully treated with topical tacrolimus ointment. Acta Derm Venereol 2012;92:104-5.
9. Takahashi S, Numakura K, Kubo K, Matsuda Y, Yamamoto R, Honma N, *et al*. A case of penile pyoderma gangrenosum treated with steroid administration without penectomy. Hinyokika Kiyo 2019;65:219-22.