

Penile pyoderma gangrenosum treated with cyclosporine: Case report

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Abstract

Pyoderma gangrenosum (PG) of the penis is a very rare entity in medicine and it can be destructive. Generally, pyoderma gangrenosum is known to be common among patients with systemic diseases such as inflammatory bowel diseases, polyarthritis, diverticulosis, paraproteinaemia, myeloma, leukaemia, active chronic hepatitis, and Behcet syndrome. Early diagnosis and administering effective treatment can prevent permanent damage to the penis and save the patient's social life. Herein we report a delayed diagnosis of Pyoderma gangrenosum of the penis resulting in partial damage and was effectively treated with cyclosporine.

Keywords: Pyoderma gangrenosum, Cyclosporine.

Introduction

Pyoderma gangrenosum is an uncommon inflammatory noninfectious skin disease characterized by heavy neutrophilic infiltration.¹ It is defined by the presence of an ulcer that is irregular in shape, wet spongy ground with raised undermined borders and purulent necrotic bases that is painful in nature.¹ It commonly affects the lower extremities as well as the buttocks, abdomen, and face.¹ Furthermore, few cases of penile PG have been reported. It is mostly idiopathic; nevertheless, underlying immunological anomalies have been recognized.² Fifty percent of the patients have underlying systemic diseases such as inflammatory bowel diseases, polyarthritis, diverticulosis, paraproteinaemia, myeloma, leukaemia, active chronic hepatitis, and Behcet syndrome.¹ There are four known variants of pyoderma gangrenosum; ulcerative, pustular, bullous and superficial.³ There are many options namely immunosuppressant medications (glucocorticoids and cyclosporine), aminosalicylate and, more recently, tumour necrosis factor blocker drugs.¹ In general, the treatment of pyoderma gangrenosum is very challenging and difficult.

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In this Case-report, we will present a patient with penile pyoderma gangrenosum who was successfully treated with cyclosporine.

Case History

A 26-year-old single man who was otherwise healthy, developed multiple painful small ulcers over the tip and shaft of his penis for few weeks. Before seeing a physician, topical creams were used without any benefit. After being seen by a dermatologist he was given oral antibiotics for 2 weeks but without any response even he got worse. In November 2015, he was referred to our clinic at King Khalid University Hospital with the already disfigured tip of the penis with the larger ulcer (Figure-1). To exclude infectious causes, swab culture was done and showed minimal growth of staphylococcus epidermidis. Moreover, a punch biopsy was taken from the penile skin which showed epidermal and dermal loss with necrotic debris overlying a granulation tissue at the base of the ulcer. There were extensive dermal inflammatory infiltrates predominantly lymphocytes, abundance of plasma cells and few neutrophils affecting the perivascular areas with one blood vessel demonstrating fibrinous necrosis that was consistent with pyoderma gangrenosum. A 300 mg oral cyclosporine was started after baseline labs came to be normal including renal and liver function tests. Significant improvement was



Figure-1: Disfigured tip of the penis with a 4×3 cm ulcer on the dorsal aspect.



Figure-2: Healing Pyoderma gangrenosum of the penis with residual damage after two months of treatment.

apparent within 2 weeks. Two months later all lesions healed completely with residual damage (Figure-2). The dose was decreased to 100 mg for 2 months without any reactivation. After few weeks of discontinuing the treatment, patient started to notice few papules for which cyclosporine was resumed at 100 mg daily and continued for 3 months then stopped. During this period patient observed one or two painful papules which appeared when he ran short of cyclosporine for more than a week. However, stopping the treatment for three months lead to complete remission. Consent was taken from the patient for publishing the report and confidentiality was ensured.

Discussion

Pyoderma gangrenosum (PG) is quite a rare idiopathic destructive skin disease. It is characterized by the progressive growing ulcer, and it commonly affects the lower extremities as well as the buttocks, abdomen, and face; however, it is more common in the former than it is in the latter ones.¹ PG rarely affects the genital area. Furthermore, less than 25 cases of penile PG have been reported. Fifty percent of the patients have underlying systemic diseases.¹ Diagnosis of PG might be challenging because many other diseases share almost the same clinical picture such as infectious, vasculitis, malignancy, insect bites, and venous or arterial insufficiency.⁴ Dermatologists should have a lower threshold of skin biopsy for any chronic ulcer of the penis. Thereby, making early diagnosis and initiate early treatment to prevent permanent damage to the penis. In our case, diagnosis and treatment were delayed for unknown reasons from the primary care physician for few weeks and another two weeks after the first dermatologist which resulted in significant

damage of the penis. Certain patients might hesitate to take a biopsy from the penis. However, with high clinical suspicion of PG even without biopsy, cyclosporine still can be initiated empirically without biopsy as a trial for treatment. The response is usually rapid, which may support the diagnosis. A short course of cyclosporine generally safe and can be given even without confirming the diagnosis if infections have been excluded. Cyclosporine safety profile can be observed from the wide use of it in children with atopic dermatitis and other diseases.⁵ We highly recommend starting cyclosporine treatment for few weeks if suspecting the case of PG of the penis in patients refusing biopsy. In our case, the patient was kept on a low dose of cyclosporine 100 mg for 3 months for complete healing as he reported few lesions every time he missed taking cyclosporine. It is crucial to follow patients after complete healing for at least 6 months to observe for possible reactivation of PG. Probable differential diagnosis would be Sweet syndrome. However, it is less likely with the histopathological findings because it mainly expresses neutrophilic cell infiltrate without clear vasculitis or ulceration which were not seen in our patient. Small vessel vasculitis is another possibility that lacks mixed inflammatory infiltrates and the minimal involvement of blood vessel. Also, both diseases differ clinically. Several therapeutic options with different response rates have been known. Corticosteroids and cyclosporine by far is considered as a gold standard treatment.⁴ Other treatment options have been also effective, which include steroids and tacrolimus, colchicine, dapsone, and thalidomide.⁶⁻⁹ Georgia et al first reported the potential use of cyclosporine in addition to corticosteroids for treating penile PG.¹⁰ Cyclosporine works in T-helper lymphocytes by inhibiting transcription of interleukin (IL)-2 and other cytokines. However, nephrotoxicity, hypertension, and neurotoxicity might develop in a long-term use.¹¹ In this case, the treatment of cyclosporine was very effective without any adverse effect.

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Conflict of Interest: None to declare.

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