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Penile Pyoderma Gangrenosum in Pediatric Ulcerative Colitis

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A 17-year-old male was hospitalized with 6 weeks of worsening lower abdominal pain associated with >10 episodes of bloody diarrhea per day and weight loss. The patient endorsed additional complaints of bleeding from a painful ulcerative lesion at the dorsal penile base. This appeared 2 weeks prior as a single pustule before subsequent rupture and ulceration. Magnetic resonance enterography identified radiological features compatible for pancolitis, consistent with histological findings on colonoscopy diagnostic for ulcerative colitis (UC). Small bowel disease involvement inclusive of the terminal ileum was excluded by unremarkable magnetic resonance enterography and esophagogastroduodenoscopy. Relative to the penile lesion, evaluation for sexually transmitted infections was negative. There were no oral, ophthalmologic, or intestinal manifestations to suggest Behçet syndrome. Dermatology consultation considered penile pyoderma gangrenosum (PG) as a leading differential diagnosis in the clinical setting of UC, pathergy at the lesion site, and ultimately the rapid therapeutic response to systemic corticosteroids and antibiotics (Fig. 1). Biopsy from the lesion site was not recommended based on the above-mentioned clinical grounds. Gastrointestinal complaints related to the patient's UC were corticosteroid responsive, with anticipation to start a biologic as an outpatient.

Cutaneous manifestations of inflammatory bowel disease (IBD) are well described with an incidence of 10%–15% in pediatric IBD.[1] PG occurs in less than 5% of children with IBD, having no association to intestinal disease activity.[2] PG is an autoinflammatory progressive ulcerative skin disorder, often associated with systemic disease. Diagnosis is made on a clinical basis and biopsy can be supportive but nonspecific.[3] Alternative causes of genital ulcerations warrant exclusion, where infectious and noninfectious conditions are known to have similar cutaneous findings. Syphilis, herpes simplex, and *Haemophilus ducreyi* comprise common infectious agents for4genital ulcerations.[3] Mycobacterial, fungal, and parasitic infections should be ruled out if relevant. Noninfectious causes encompass a wide differential including but not limited to squamous cell carcinoma, Behçet syndrome, ulcerative sarcoidosis, antiphospholipid syndrome, Sweet syndrome, and metastatic Crohn disease.[4] Clinical presentation of PG typically involves a single or numerous pustules that rapidly progress to painful ulcerations typically on the extremities, head, or gluteal region in children.[4] Genital involvement of PG is rare, limited to adult case reports with the majority involving underlying Crohn disease and a single adult case of penile PG associated with UC.[5] Management is to treat the underlying systemic disease, and includes systemic and local immunosuppression with emphasis on wound care and pain control.[6]

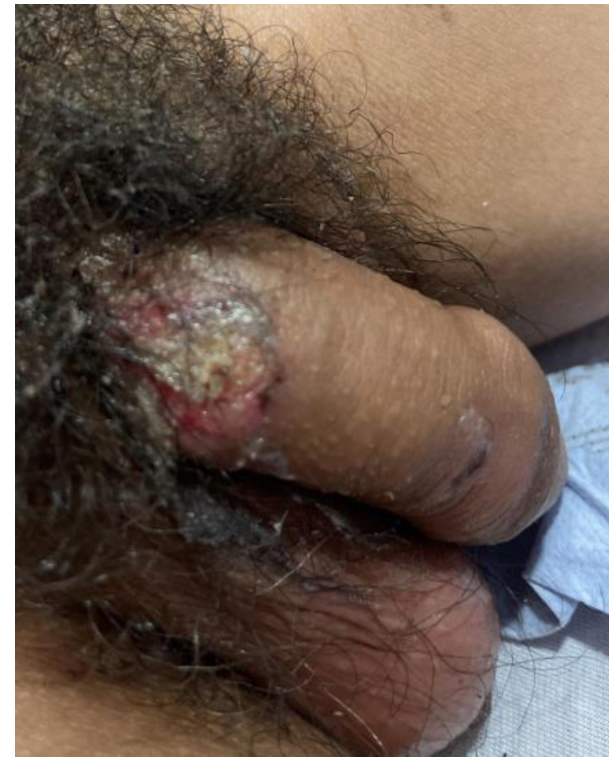

FIGURE 1. Dorsal penile base ulcerative lesion, superficial base with scab formation post 2 d of systemic corticosteroids.

This is one of the first reports of a case of penile PG associated with pediatric UC. Clinicians should be aware of these unique presentations of cutaneous manifestations linked to IBD.

ACKNOWLEDGMENTS

Informed consent from the patient and their parent was obtained prior to publication of the case details.

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Received August 23, 2022; accepted November 12, 2022.

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The authors report no conflicts of interest.

JPGN Reports (2023) 4:1(e281)
ISSN: 2691-171X
DOI: 10.1097/PG9.0000000000000281

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