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Vaginal cuff pyoderma gangrenosum with associated ureteral stricture: A case report

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ABSTRACT

Pyoderma gangrenosum is a sterile inflammatory disease of unknown etiology characterized by recurrent cutaneous ulcers. It can occur in extracutaneous locations, especially at operative sites, and has been reported following gynecologic surgery. This report is the first case of pyoderma gangrenosum as a remote complication of pelvic surgery with associated ureteral stricture. It demonstrates the diagnostic challenge of this rare disease and the importance of broadening the differential diagnosis when apparent infections do not respond to treatment to minimize the morbidity of ineffective antibiotic and surgical interventions.

1. Introduction

Pyoderma gangrenosum (PG) is a sterile neutrophilic dermatosis characterized by recurrent cutaneous ulceration; it is often associated with inflammatory bowel disease, rhematic disorders, or neoplasms.[1] Although rare, there are gynecologic reports of PG.[2,3] To the best of our knowledge, this report is the first case of PG arising as a remote complication of pelvic surgery that also involves adjacent urologic organs.

2. Case presentation

A sixty-year-old woman with surgical history pertinent for vaginal hysterectomy twenty years ago and sacrospinous ligament suspension fifteen years ago presented to the urology clinic for evaluation of vaginal discharge associated with right hydronephrosis. Symptoms began ten months prior to presentation when she developed dyspareunia followed by vaginal bleeding and discharge. She then developed low-grade fevers, chills, fatigue, poor appetite, weight loss, and weakness. No dysuria, gross hematuria, flank pain, or suprapubic pain.

A gynecologist performed a pelvic exam and removed a right vaginal suture that appeared infected. She subsequently completed three courses of oral antibiotics without resolution of symptoms. Nine months after symptom onset, vaginal culture grew Bacteroides and MRI was obtained showing new-onset hydronephrosis associated with fluid collection at vaginal cuff (Fig. 1). Renal function was normal. Oral levofloxacin and metronidazole were started and switched to intravenous due to side effects. She then developed scattered skin lesions.

Medical history includes hypertension, hyperlipidemia, hypothyroidism, mood disorder, and psoriasis treated with ixekizumab (humanized IgG4 monoclonal antibody specific for interleukin 17A). No personal or family history of inflammatory bowel disease, rheumatoid arthritis, or malignancy. She is a G5P4A1 with all spontaneous vaginal deliveries. She is a non-smoker.

Physical exam revealed scattered violaceous papules across her face and multiple pustular nodules of the distal extremities concerning for septic emboli (Fig. 2). Dried purulent discharge was noted on otherwise normal genitalia. Pelvic exam revealed an open vaginal cuff with intact perineum and copious purulent material.

The patient was admitted with concern for subacute bacterial endocarditis. Vaginal and blood cultures were negative. Finger lesions underwent incision and drainage; wound cultures were negative. Cystoscopy with right retrograde pyelogram showed a draining vaginal cuff fluid collection, bladder inflammation, and right hydroureteronephrosis to the level of the bladder. Three vaginal cuff sutures (appearance most consistent with braided polyester) were removed. Vaginal cuff tissue cultures were negative. Additional soft tissue lesions developed despite broad antibiotic coverage. Echocardiography and autoimmune studies were unremarkable. Dermatology was consulted and expressed high suspicion for PG given history and exhaustive negative infectious work-up. Punch biopsy was performed demonstrating a neutrophilic infiltrate supportive of the diagnosis. The patient

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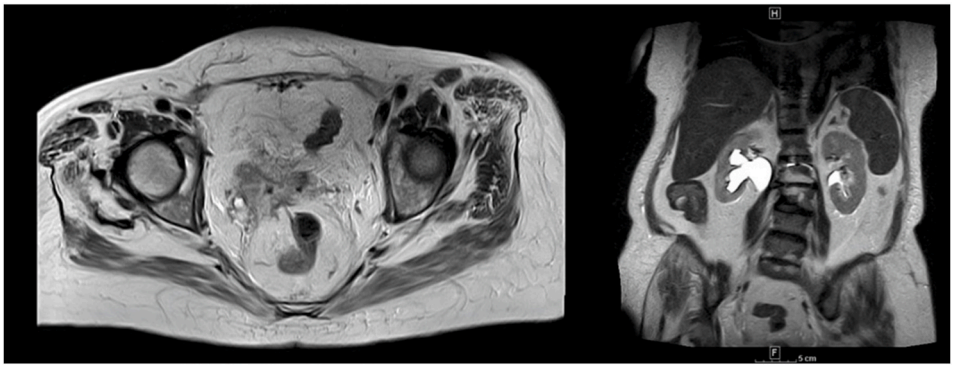

Fig. 1. MRI depicting right hydroureteronephrosis associated with right vaginal cuff fluid collection.

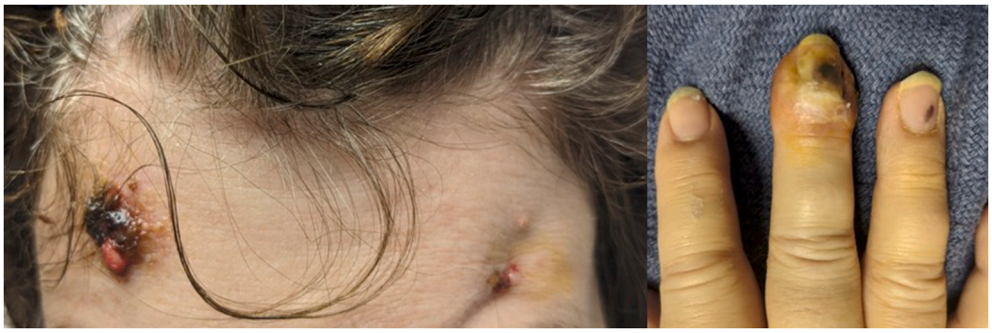

Fig. 2. Representative lesions on face and distal extremities at time of presentation to urology clinic.

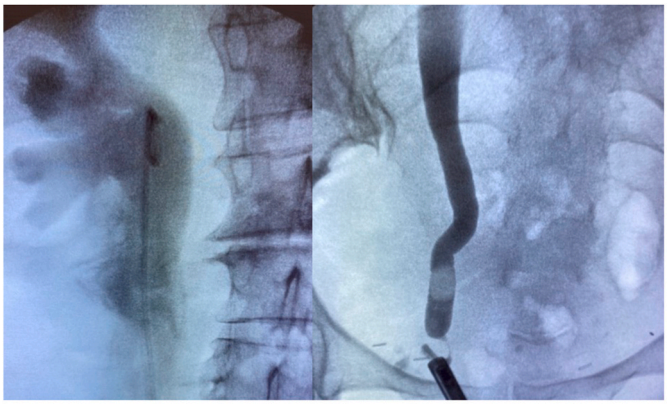

Fig. 3. Right hydroureteronephrosis to the level of the bladder with severe distal ureteral stricture associated with severe vaginal inflammation.

was initiated on cyclosporine with rapid response of skin lesions.

One month later, she had persistent vaginal discharge and right hydronephrosis with normal renal function despite overall good response to cyclosporine. Repeat cystoscopy with right retrograde pyelogram demonstrated vaginal cuff inflammation, severe hydroureteronephrosis, and distal ureteral stricture that was stented (Fig. 3). The patient was transitioned to adalimumab for long-term management. Her ureteral stricture is currently managed with ureteral stent exchanges. When her PG is adequately controlled and her vaginal cuff has healed, she will undergo ureteroneocystostomy.

3. Discussion

Onset of PG typically occurs in the third to sixth decade of life with a higher incidence in women. This rare disorder usually presents on the lower extremities as painful pustules that rapidly enlarge into ulcers with violaceous undermined borders, however, it can occur in other cutaneous or extracutaneous location. Etiology of PG is unknown but involves aberrant autoimmune response to unspecified antigen(s). With no definitive diagnostic criteria, it is a diagnosis of exclusion. Differential diagnosis includes vascular occlusive disease, vasculitis, malignancy, infectious diseases, tissue injury, and drug reactions. Diagnosis is supported by response to treatment, typically systemic corticosteroids or cyclosporine.[1]

Gynecologic reports of PG are rare. One report describes chronic ulceration of the vagina following cervical cone biopsy.[2] Another case occurred in the vaginal vault as a late complication (post-op day 92) of vaginal hysterectomy.[3] More commonly post-operative PG arises at surgical incisions during the immediate post-operative period, with breast and abdomen being the most common sites.[4] In post-operative PG, trauma upregulates neutrophils at the site of ulceration. The role of suture material is unknown. It has been theorized that higher reactivity material (eg., silk) with epidermal puncture is more likely to elicit PG, however, no prospective data exists to support this theory and many cases occur after low reactivity absorbable subcuticular sutures are employed.[4] Similarly, one case report found no difference in PG lesions created by seven different suture types.[5] In this case braided polyester, a low reactivity non-absorbable suture, was identified. Furthermore, post-operative PG has a lower association with system disease (34 vs 50%) compared with classic PG; most patients with post-operative PG receive antibiotics and undergo wound debridement prior to diagnosis.[4]

The case presented here demonstrates the challenge of diagnosing PG in extracutaneous locations, especially by clinicians unfamiliar with this rare disease. It highlights the importance of considering alternative etiologies when seemingly infectious problems do not respond to antibiotic therapy. Furthermore, the remote history of pelvic surgery suggests foreign bodies can serve as a source of pathergy in PG beyond the acute peri-operative period. Involvement of adjacent urologic organs in this case add an additional layer of complexity. Ureteral stricture

secondary to inflammation from PG has created a treatment conundrum where only additional operative trauma during ureteroneocystostomy can definitively resolve the consequence of the initial disease process.

4. Conclusion

This case is the first report of PG of the vaginal cuff arising in a patient with a remote history of pelvic surgery; the involvement of adjacent urologic organs is also distinctive. It demonstrates the diagnostic challenge of this rare disease and underlies the importance of maintaining a broad differential diagnosis when apparent surgical infections do not respond to treatment. Early consultation of a dermatologist if concerned for PG can minimize the morbidity of repetitive antibiotic and surgical interventions that do not effectively treat this disease.

Consent

Informed consent was obtained from the patient to include case details in this written report.

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