

Traumatic vulvar aphthous ulcers: a case series

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Keywords: Vulvar ulcer, aphthous, pyoderma gangrenosum

Abstract

We present three women who each presented with a vulvar ulcer consistent with aphthous. In each case a biopsy was performed which excluded malignancy and revealed non-specific findings. All women responded following intralesional steroid injections. We initially considered a diagnosis of pyoderma gangrenosum. However, the presentation and clinical findings were more consistent with aphthous ulcer.

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infectious etiology, a biopsy can also give information regarding potential non-infectious etiologies such as dermatoses, immunobullous disorders, systemic diseases, and malignancy.¹ Here we present three women with vulvar pain and exam notable for a single vulvar ulcer; with history significant for no associated genitourinary disease, but with multiple comorbidities including morbid obesity. Each had a non-specific biopsy demonstrating a dense neutrophilic infiltrate. IRB approval is not required for case series involving less than six cases in our institution.

Background

Vulvar ulcers may be caused by infectious as well as non-infectious etiologies (e.g. aphthous, Behçet disease, pyoderma gangrenosum, malignancy). Clinically, when a patient presents with vulvar ulcer, it is important to identify and treat infectious diseases that may be the cause. In addition to a thorough history, physical exam and appropriate testing for an underlying

Cases

Patient A: a 55 year old non-Hispanic white female with a past medical history significant for chronic neck pain treated with steroids and narcotics, hypertension, and body mass index (BMI) 50, presented with vulvar burning, itching, and ulcer. Prior evaluation for the cause of ulceration was negative including herpes simplex virus PCR and serology, syphilis serology, and human

Please cite this paper as: Pierick AR, Elas D, Stockdale CK. Traumatic vulvar aphthous ulcers: a case series. Proc Obstet Gynecol. 2017;7(3): Article 3 [6 p.]. Available from: <http://ir.uiowa.edu/pog/> Free full text article.

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Financial Disclosure: The authors report no conflict of interest.

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immunodeficiency virus (HIV) serology. Vulvar exam disclosed a well demarcated 2.5x2 cm right vestibular ulcer with rolled edges (Photo 1), and mild erythema of the intertriginous areas, pannus, and groin folds. Biopsy of the ulcer demonstrated squamous mucosa with marked acute and chronic inflammation, ulceration, granulation,

and mucinous metaplasia. Pathergy (increased ulcer size) was noted following biopsy. She received 4 triamcinolone injections (10 mg diluted to 1cc with normal saline) into the dermal edge of the ulcer over 5 months with complete resolution and re-epithelialization of the ulcer.



Photo 1: Patient A – pre treatment

Patient B: a 77 year old female with a past medical history significant for renal insufficiency, diabetes, heart failure, coronary artery disease, atrial fibrillation myocardial infarction, chronic obstructive pulmonary disease,

hypertension, hyperlipidemia, deep vein thrombosis/pulmonary embolism, ulcerative colitis, rheumatoid arthritis, vitamin D deficiency, genital herpes simplex virus (on acyclovir suppression), and BMI 51, presented with vulvar

burning and ulcer. Prior serology testing for syphilis and human immunodeficiency virus (HIV) were both negative. Additionally, previous biopsy of a well demarcated 2x3 cm left inferior vestibular ulcer with rolled edges and a clean base (Photo 2) demonstrated active granulation tissue with acute and chronic inflammation. Concern for

possible pathergy was reported following biopsy; however, the patient was unable to confirm due to inability to visualize her vulva. She received 4 triamcinolone injections (10 mg diluted to 1cc with normal saline) into the dermal edge of the ulcer over 4 months with complete resolution and re-epithelialization of the ulcer.



Photo 2: Patient B – pre treatment

Patient C: a 72 year old female living in assisted-living with past medical history significant for hypertension, coronary artery disease (required stent placement while undergoing treatment for vulvar ulcer), thyroid disease, and BMI 53, presented with vulvar burning, pain and ulcer. Prior evaluation for the cause of ulceration was negative including

herpes simplex virus PCR and serology, syphilis serology, and HIV serology. Previous treatments included fluconazole, clotrimazole, clobetasol, corticosteroid suppositories, and topical lidocaine without improvement of symptoms or the ulcer. Exam disclosed a well demarcated 3x2 cm superficial ulcer with rolled edges and clean base

at the left superior vestibule. Biopsy demonstrated squamous mucosa with chronic inflammation. Pathergy was noted following biopsy. She received 10 triamcinolone injections (10 mg diluted to 1cc with normal saline) into the dermal edge of the ulcer over 10 months with complete resolution of the ulcer. She had a minor set-back with return of pain and superficial ulcer three months after the initial resolution. Repeat biopsy demonstrated squamous mucosal ulceration. The ulcer resolved with attention to vulvar hygiene including peri-wash, zinc oxide ointment barrier, and avoidance of friction including mechanical irritation.

Discussion

We present three cases of well demarcated superficial vulvar ulcers with rolled edges responsive to intralesional steroids. Biopsy of the

ulcers demonstrated nonspecific findings.

While we initially considered a diagnosis of pyoderma gangrenosum especially in light of the concern for pathergy following biopsies, the clinical presentation was not consistent with pyoderma gangrenosum, which includes ulcer with undermined livid (bluish) borders or pyodermic-like erosions with surrounding erythema.² Furthermore, diagnostic criteria for pyoderma gangrenosum require two major criteria and at least two additional criteria (Table 1).^{2,3} However, none of the cases presented involved an ulcer with a livid, undermined border. Thus, while all of the cases presented had several of the additional criteria, a diagnosis of pyoderma gangrenosum requires both major criteria as well as 2 additional criteria.

Table 1: Diagnostic criteria for pyoderma gangrenosum^{2,3}

Definitive diagnosis requires 2 major and at least 2 additional criteria	
Major criteria:	Primary sterile pustule or ulcer with livid, undermined border, AND
	Exclusion of other relevant differential diagnoses such as chronic venous/arterial leg ulcer, pyodermatitis, vasculitis
Additional criteria:	Histology of the wound border: neutrophil infiltration of the dermis with signs of vasculitis and accumulation of immunoglobulins and/or complement factors
	Existence of relevant, associated concomitant diseases
	Response to a conventional ulcer therapy
	Triggering of pyoderma gangrenosum by pathergy phenomenon
	Extremely painful ulcer

Adapted from Wollina R. Pyoderma gangrenosum – a systemic disease? Clinics in Dermat 2015;33:527-530; Al Ghazal P, Herberger K, Schaller J, Strölin A, Hoff NP, Goerge T, Roth H, Rabe E, Karrer S, Renner R, Maschke J, Horn T, Hepp J, Eming S, Wollina U, Zutt M, Sick I, Splieth B, Dill D, Klode J, Dissemond J. Associated factors and comorbidities in patients with pyoderma gangrenosum in Germany: a retrospective multicentric analysis in 259 patients. Orphanet J Rare Dis. 2013 Sep 8;8:136.

We believe the etiology of the ulcers in the women presented to be aphthous ulcers of unknown etiology. As noted, all three cases involved morbidly obese older women with multiple chronic medical co-morbidities. They all experienced similar symptoms of vulvar pain and itching made worse with sitting. All were treated with medications primarily directed at various infectious etiologies without improvement prior to referral to our institution. The cases described appear of similar etiology and responded to intralesional steroid injections over the course of months. While many conditions (known and unknown) respond to steroids, the similar history and clinical features and response in each case suggests a similar etiology. Other diagnostic considerations included malignancy as well as Behçet's disease, a chronic relapsing systemic vasculitis. As with pyoderma gangrenosum, the diagnosis of Behçet's disease is based on clinical criteria (including: recurrent oral ulceration, minor aphthous, major aphthous, herpetiform ulceration that has recurred at least three times during a 12 month time period; plus two additional criteria: recurrent genital ulceration, eye lesion, skin lesions, pathergy).⁴ Thus, none of the women presented had criteria to suggest Behçet's disease and biopsies excluded malignancy. Dermatoses were also excluded based on clinical examination and biopsy findings (e.g. lichen planus, lichen sclerosus) as well as exclusion of immunobullous disorders. One patient noted a past medical history including ulcerative colitis. While inflammatory bowel disease (IBD) is associated with vulvar ulceration, the clinical and biopsy

findings were not supportive for IBD.

Thus, our leading diagnosis for each of the patients presented is aphthosis ulcer; an uncommon, nonsexually transmitted condition characterized by painful, necrotic ulcerations of the vulva or lower vagina.⁵ Similar to oral canker sores, the classic description includes ulceration with a well-defined margin and a rolled erythematous border; which can be large (up to 3 cm).⁵ Well known inciting factors include trauma.⁶

We theorize that some small trauma (such as that caused by wiping or fingernail injury) caused injury to the vulvar mucosa and was the inciting event for each of the cases presented. The vulva and vestibule are vulnerable to irritation because the barrier function is weaker than other skin sites due to differences in structure, occlusion, and moisture.⁷ Minor trauma happens regularly, but in these patients the area was especially prone to mechanical friction/ trauma and likely unable to heal appropriately due to a combination of impaired mucosal perfusion, and probable chronic irritation from friction/rubbing and/or immunosuppression from associated comorbidities including morbid obesity. Furthermore, it is difficult to maintain appropriate vulvar hygiene in obese patients with limited mobility, which can contribute to impaired barrier function. With steroid injections and guidance regarding hygiene, all three patients responded.

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