Botryomycosis of the Vulva: A Case Report

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■ **Abstract**

Objective. Vulvar ulcers may be caused by various etiologies including infection, trauma, dermatosis, and cancer. We report a case of a vulvar ulcer caused by botryomycosis.

Case. An 85-year-old woman presented with vulvar itching, pain, bleeding, and ulcerations suspicious for cancer. Biopsies of the ulcers returned without dysplasia or malignancy. She was referred to the tertiary care vulvar vaginal disease clinic. Dermatopathologic reevaluation of pathologic slides diagnosed lichen sclerosus and botryomycosis. The patient was treated with ciprofloxacin for 7 weeks with complete resolution of vulvar ulcerations.

Conclusion. Botryomycosis should be included in the differential diagnosis of infectious etiology of vulvar ulcers. ■

Key Words: botryomycosis, vulvar ulcer

Genital ulcerations may have vastly different etiologies, yet they have very similar clinical presentation. When a patient presents with a genital ulcer, the clinician is given the challenge to accurately diagnose the etiology of the ulcer and provide the appropriate treatment. Common causes of genital ulcers include infection, trauma, dermatosis, aphthous ulcers, and malignancy.

Botryomycosis is an uncommon bacterial inflammation of the soft tissue, bone, and viscera. It is characterized by chronic, purulent, and granulomatous inflammation. Common causative organisms of botryomycosis include *Staphylococcus aureus*, *Pseudomonas aeruginosa*, *Escherichia coli*, *Streptococcus* sp, and *Proteus* sp. The histologic hallmark is the presence of granules in a suppurative focus. Initially, this disease process was thought to be fungal in origin and was given the Greek-derived name botryomycosis: *botryo* meaning bunch of grapes and *mycosis* meaning fungal in origin. This is a misnomer because it was later determined that this condition was bacterial in origin. Its exact pathogenesis is poorly understood. It has been associated with immunocompromised patients rather than a virulent strain of the infecting bacteria. Without proper treatment, prognosis is poor.

This is the first case of cutaneous vulvar botryomycosis reported in the literature.

CASE REPORT

In January 2012, an 85-year-old, G2P2, woman presented to her local gynecologist with a 5-week history of intermittent vulvar bleeding, sores, and itching. Vulvar examination documented right and left periclitoral ulcers. The initial clinical impression was suspicious for vulvar cancer. Three vulvar biopsies were obtained: 2 from the right ulcer and 1 from the left ulcer. Local pathology returned acute inflammation and lichen simplex chronicus with ulceration negative for neoplasia. The patient was immediately referred to our tertiary care center vulvar vaginal disease clinic for evaluation. The patient stated she had a long-standing history of severe vulvar itching and felt she "may have torn" herself with scratching before her bleeding started. She was postmenopausal for 37 years, no previous history of abnormal cervical cytology, no gynecologic surgeries, no history of hormone replacement, and not sexually

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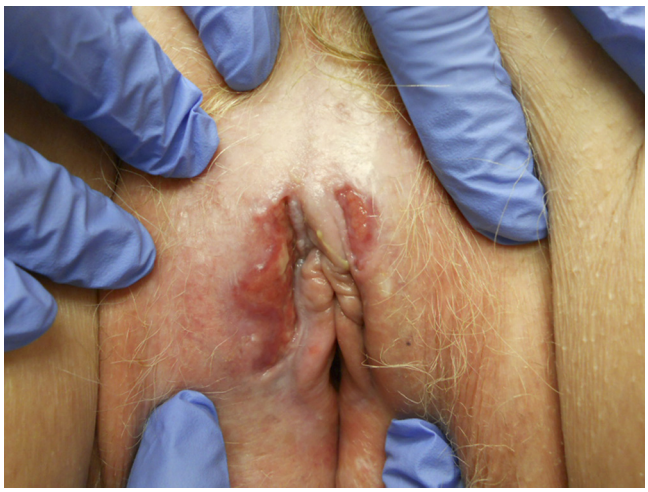

Figure 1. Initial presentation: vulvar erosion/ulcers.

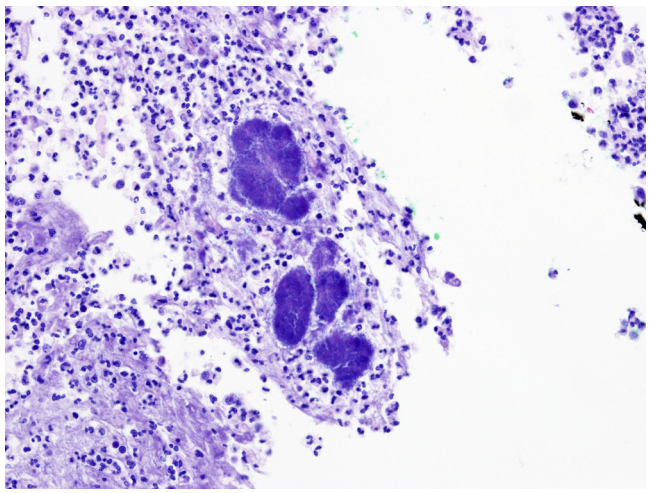

Figure 3. Basophilic granules with eosinophilic zonation and surrounding suppurative inflammation (hematoxylin and eosin, ×400).

active. On vulvar examination, she was found to have clinical changes suggestive of lichen sclerosus as well as atrophic vaginitis. The patient had extensive erosive and deep ulceration involving the bilateral inner labial sulci lateral to the clitoris (see Figure 1).

The original vulvar biopsy slides were reviewed by our dermatopathologists (B.S., M.S.). Diagnosis from this review returned as lichen sclerosus with associated infection with gram-positive cocci in clusters suggestive of *Staphylococcus*. The presence of bacteria in grains is consistent with botryomycosis. Hematoxylin and eosin–stained sections demonstrated an acanthotic epidermis with underlying dermal hyalinization and scant band-like lymphoid inflammation. In addition, there was a subepidermal suppurative microabscess with associated basophilic granules with eosinophilic zonation (see Figures 2 and 3). High-power examination revealed the basophilic material to be composed of numerous gram-positive cocci in clusters and focally in chains (see Figure 4). No fungal forms were seen on Gomori methenamine silver-stained sections.

After a review of the literature, the patient was empirically treated orally with ciprofloxacin 250 mg twice a day and sulfamethizole/trimethoprim DS twice a day. She was also given oral fluconazole 200 mg for yeast prophylaxis. Lesional cultures obtained returned growing 20% to 30% Group B *Streptococcus* sensitive to penicillin and ampicillin and 60% to 70% *E. coli* sensitive to ciprofloxacin,

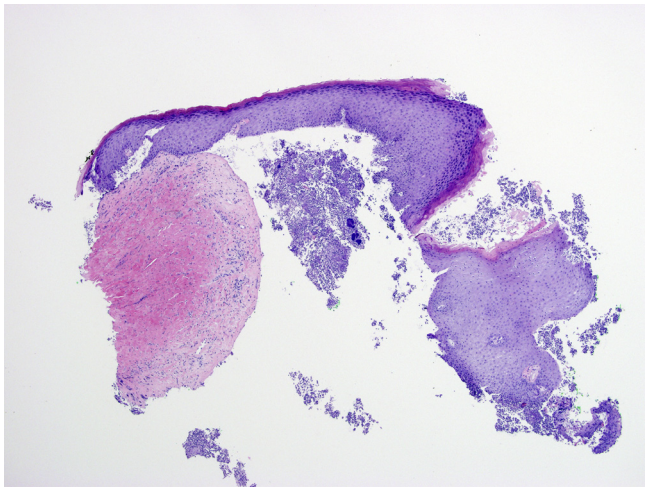

Figure 2. Dermal sclerosus with associated suppurative microabscess and basophilic granules in the superficial dermis (hematoxylin and eosin, ×40).

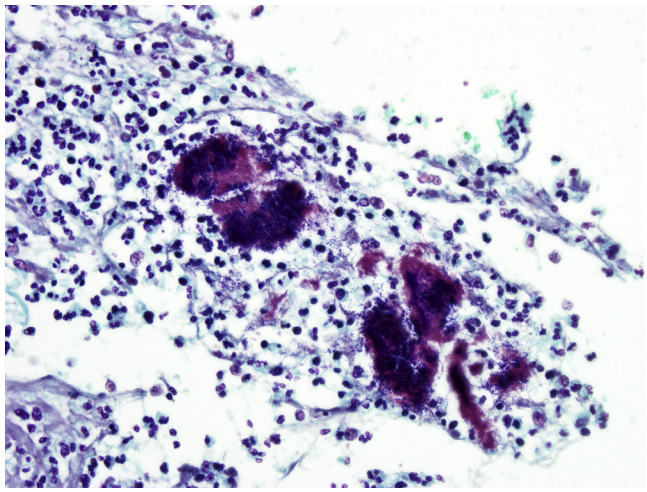

Figure 4. Botryomycotic granules demonstrating gram-positive cocci consistent with *Staphylococcus aureus* (Gram stain, ×600).

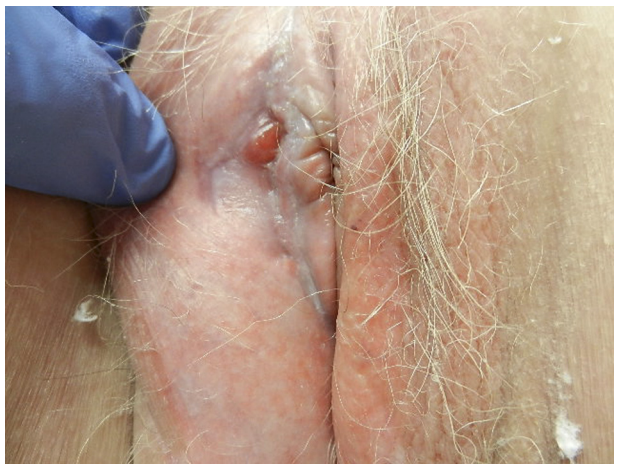

Figure 5. Four-week follow-up: resolution of erosion/ulcers.

levofloxacin, cefazolin, or gentamicin. She discontinued sulfamethizole/trimethoprim DS because of gastric intolerance after 7 days, whereas she continued ciprofloxacin 250 mg twice a day. The patient was also treated with supportive measures of aluminum acetate 1:40 solution compressed to the area daily, zinc oxide ointment 3 times a day as a skin protectant to the area, and strict adherence to vulvar hygiene to avoid chemical and mechanical irritation.

The patient returned after 4 weeks with resolution of her symptoms of itching and bleeding. Clinically, the left ulceration was resolved and the right ulcer was now a 6-mm area of granulated tissue (see Figure 5). The patient continued ciprofloxacin 250 mg twice a day for 4 weeks, conjugated estrogen vaginal cream, and zinc oxide ointment 1 to 2 times a day as a skin protectant and continued strict adherence to vulvar hygiene care. At her 7-week follow-up, she continued to be asymptomatic and the ulcerations were completely healed (see Figure 6). At the 1-year follow-up, she had no further vulvar/vaginal problems.

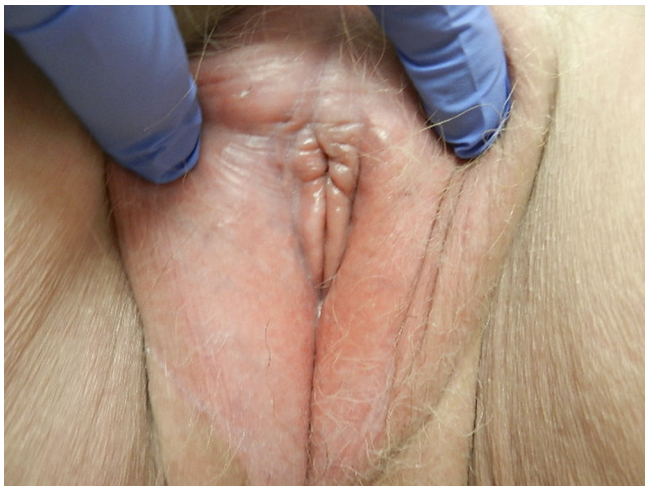

Figure 6. Seven-week follow-up: resolution of erosion/ulcers.

DISCUSSION

Botryomycosis is a long-term bacterial disease that clinically and histopathologically resembles a fungal infection first described in 1870 by Bollinger as fungus-like granules occurring in the lungs of a horse [1]. The term *botryomycosis* was eventually used because of the grape-like (*botryo*) appearance of the grains and the presumed fungal (*mycosis*) origin [1, 2]. Because the infection is bacterial in origin and not mycotic, other terms have been used to describe it as well, including actinophytosis, bacterial pseudomycosis, granular bacteriosis, and staphylococcic actinophytosis [3].

Botryomycosis in humans was first documented in 1913 by Dr Eugene Opie on an autopsy of the liver of an 11-year-old girl [4]. Since then, case reports of cutaneous, bone, and visceral botryomycosis have been presented in the literature as unusual causes for bacterial infections to be considered in differential diagnosis.

Botryomycosis in humans is most often associated with *S. aureus* infection [3]. Other implicated bacteria include *P. aeruginosa*, *E. coli*, *Bacteroides* sp, *Proteus* sp, *Streptococcus*, *Bacillus* sp, *Actinobacillus lignieresii*, and *Propionibacterium acnes* [5, 6]. Clinically, botryomycosis is divided into visceral and cutaneous types [1, 2]. Cutaneous botryomycosis is most common and presents with single or multiple inflammatory nodules, abscesses, ulcers, sinuses, and fistulas that drain microscopic small white granules [2, 5]. Frequent sites of involvement include the extremities, especially sites of trauma such as the hands and feet, as well as the head and neck area and inguinal and gluteal areas [5, 6].

Histopathologically, botryomycosis is characterized by granules, resembling those of actinomycosis, ranging in size from a few micrometers to 2 mm in diameter [5]. These granules are composed of aggregates of nonfilamentous bacteria with radial deposition of PAS-positive eosinophilic material containing immunoglobulins, representing the so-called Splendore-Hoeppli phenomenon, and are associated with surrounding suppurative inflammation and granulation tissue [5, 7]. Gram-stained sections are useful to highlight the causative bacterial organism combined with microbiologic culture studies for the isolation of the definitive agent and to determine the antibiotic sensitivity patterns.

The exact pathogenesis of botryomycosis is not well understood. However, the condition is often associated with a defect in either local or systemic host resistance [6]. Predisposing systemic conditions include general debilitation, diabetes mellitus, liver disease, cystic fibrosis, alcoholism, malnutrition, and use of systemic corticosteroids [6, 8]. Cutaneous disease has been associated with trauma, foreign bodies, topical corticosteroids, and intralesional corticosteroids [6, 8]. In our patient, the combination of advanced age and trauma from scratching the untreated vulvar atrophy, lichen sclerosus, and resultant lichen simplex chronicus likely represents the underlying local immune defect that predisposed her to develop this infection.

To our knowledge, this is the first case of vulvar botryomycosis. Accurate diagnosis was made histologically with the expertise in dermatopathology and wound culture. Appropriate antibiotic therapy resulted in resolution of the erosions and ulcers. This bacterial infection should be considered as a differential diagnosis in females who present with chronic suppurative, granulomatous, or ulcerative lesions.

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