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CASE REPORT

Acute genital ulceration in an adolescent

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Abstract

Acute genital ulcer, also known as Lipschütz ulcer, is an uncommon, non-sexually transmitted condition characterized by the rapid onset of a necrotic and painful ulcer in the vulvar region. It occurs in adolescents and young women, with or without sexual activity. It may be preceded by influenza-like or mononucleosis-like symptoms. Acute genital ulcers have been associated with Epstein-Barr virus (EBV) infection or other viral or bacterial infections. However, in most cases, the cause is not determined. We report the clinical case of an 11-year-old girl without previous vaginal or oral ulceration without sexual contact who sought medical assistance with a rapidly progressive vulvar ulcer wich had started six days before, preceded by episodic low fever. The exclusion of infectious causes, the spontaneous healing of the lesion and the indeterminate serology for Epstein-Barr led to the diagnosis of ulcer of Lipschütz.

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INTRODUCTION

Diagnosing genital ulcerations in the pediatric population may be challenging, since most cases are not caused by sexually transmitted infections (STIs). The appearance of genital ulcerations may cause considerable anxiety in adolescents and their families and raise questions about whether they are sexually active, STIs, and suspicions of sexual abuse.

CASE REPORT

An 11-year-old adolescent without a relevant personal or family history of disease involving recurrent oral or genital ulcerations, skin lesions or exanthem, arthralgia, upper airway infection, ocular inflammation, or known sexual contact was taken to the Santa Casa de Montes Claros Hospital with injuries in the vulvar mucosa. A single lesion was found initially to the left of the vulva, but on Day 4 a second lesion appeared on the right side in a symmetrical position (Figure 1). The ulcerations had well-defined painful purple borders involving the perineum bilaterally. They grew rapidly and on Day 6 the ulcerations were deeper, with extensive necrosis and fibrin on the surface. On Day 10, the lesions began to heal and on Day 19 they had healed fully. The patient had episodes of low fever five days prior to the onset of the ulcerations. She was in good general condition and without other signs or symptoms.

Test results: Epstein Barr virus: positive for IgG (78.74) and indeterminate for IgM (0.97) - close to a positive result (0,99); VDRL: negative; HIV 1 and 2: negative; ANA: negative; toxoplasmosis: negative for IgM and IgG; cytomegalovirus: positive for IgG and negative for IgM; HBsAg: negative; herpes simplex 1 and 2: negative for IgG and IgM; blood culture: no bacterial growth; complete blood count: no alterations; CRP: 24 mg/L; rheumatoid factor: negative; anti-streptolysin O: < 200 U/ml; routine urine: no alterations; Gram staining: no bacteria; urine culture: no bacterial growth; lesion swab test: no bacterial growth; ultrasound examination of the abdomen and pelvis ad computed tomography scans of the pelvis: evidence of mild hepatomegaly and absence of extension of the ulcerations into deeper planes. The ulcerations were not biopsied.

Treatment included debridement of the lesions, cleaning with saline solution, placement of an indwelling urinary catheter to alleviate pain during micturition, and analgesics. Broad spectrum antibiotics were prescribed until etiology was defined. The ulcerations improved spontaneously, with gradual healing with granulation tissue, and significant decrease of the ulcerations within 15 days.

DISCUSSION

Acute genital ulceration, also known as Lipschütz ulcer, is an uncommon, non-sexually transmitted condition characterized by the rapid onset of painful, necrotic ulcerations in the vulvar area. The ulcerations are usually large (greater than 1 cm) and deep, with purple well-defined borders and a necrotic base covered with grayish exudate. It is more commonly located on the labia minora, although it might extend onto the labia majora, perineum, vestibule, and vaginal introitus. Partially symmetrical ulcerations called kissing lesions are characteristically observed. Associated signs include labial edema and inguinal adenopathy[1-3].

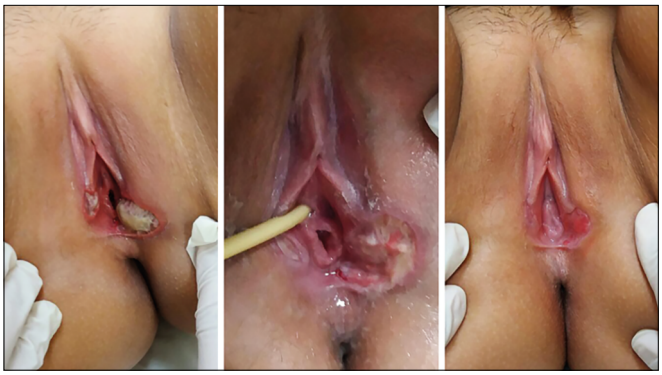


Figure 1. Evolution of the ulcerations. Chronological evolution of the ulcerations, left to right.

Acute genital ulceration is a self-limiting condition. Spontaneous healing occurs within two to six weeks of the appearance of lesions. Scarring sequelae and distortions usually do not occur (except for larger lesions). Lesions usually do not recur[13].

It typically affects adolescents and young women and is unrelated to sexual activity. It may be preceded by influenza-like or mononucleosis-like symptoms such as fever, malaise, tonsillitis, odynophagia, adenopathy, myalgia, headaches, and increased liver transaminases[1,4,7]. Acute genital ulceration has been associated with acute infection by the Epstein-Barr virus (EBV) and other viruses and bacteria such as cytomegalovirus (CMV), *Mycoplasma pneumoniae*, influenza A, among others[4,8-10]. However, in most cases the pathogen and cause cannot be determined[5,6].

The exact pathogenesis of acute genital ulceration is still unclear. A tentative explanation suggests that ulcerations are a clinical manifestation of a hypersensitivity reaction to viral/bacterial infection, with deposition of immune complexes in dermal vessels, complement activation, microthrombosis, and subsequent tissue necrosis[9,11].

Diagnosis is based on clinical signs and symptoms and exclusion and a combination of thorough patient history analysis and physical examination. Particular attention must be given to systemic conditions and ocular, neurologic, gastrointestinal, and genitourinary symptoms. Collection of information about sexual activity must be performed with the utmost privacy. Clinical criteria are described in Table 1.

The following lab tests must be ordered: viral culture, PCR, and direct immunofluorescence for the herpes simplex virus (lesion swab), since this is the most common cause of genital ulcerations. Microbiology and serology tests for other STIs are required in case of suspicion. Serology for EBV is also indicated[8-10].

Table 1. Clinical diagnostic criteria for Lipschütz ulcers in adolescents and young women.

Age < 20 years
First episode of acute genital ulceration
Absence of sexual contact or history of sexual activity in the three months prior to the appearance of ulcers
Recent history of systemic disease, influenza-like, or mononucleosis-like symptoms
Presence of one or multiple painful deep and broad vulvar ulcers with well-defined borders and a necrotic base
Bilateral kissing lesions
Acute onset with spontaneous healing occurring within six weeks
Rule out immunodeficiency
Rule out other possible causes of vulvar ulcers (diagnosis by exclusion)

Biopsy specimen examination is usually not helpful in the diagnosis of acute genital ulceration, since it produces nonspecific results such as epithelial necrosis and polymorphonuclear infiltrate. It is indicated when specific conditions are targeted[12].

Differential diagnosis includes STIs, non-infectious diseases, immune-mediated conditions (mainly Behçet's disease and inflammatory bowel disease with extraintestinal symptoms). Since some of these conditions have a late onset, patients with Lipschütz ulcers must be followed for longer for purposes of diagnostic confirmation[13,14].

Since it heals spontaneously, the treatment for acute genital ulceration is based on the management of symptoms, including reassuring the patient and her family, providing for local hygiene, wound care, and pain management[13]. Potential medications include topical and oral analgesics. Patients with severe pain and secondary urinary retention may require hospitalization, parenteral analgesia, and the placement of a urinary catheter. Systemic antibiotics must be administered to patients suspected with bacterial superinfection or vulvar cellulitis. Empirical therapy with antibiotics may also be used[13-15].

The epidemiology of the patient and the evolution of her vulvar ulcer, particularly the bilateral symmetrical appearance of kissing lesions, suggested a diagnosis of Lipschütz ulcer. Serology suggestive of acute EBV infection supported the diagnosis, since this is the pathogen that triggered the hypersensitivity reaction.

CONCLUSION

Lipschütz ulcer is diagnosed based on clinical findings and by exclusion. It is often disregarded in differential diagnosis for vaginal ulcerations in sexually inactive adolescents and young women, thereby delaying diagnosis and leading to unnecessary testing and treatment and causing physical and psychosocial pain to patients and their families.

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