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VULVAR APHTOSIS IN AN ADOLESCENT GIRL: A CASE REPORT

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ÖZET

Giriş: Vulvar aftozis patogenezinde; Epstein-Barr virüs, Sitomegaolovirüs ve İnfluenza A gibi çok çeşitli viral enfeksiyonlar sorumlu tutulmuştur. Bu yazıda vulvar aftozis tanısı alan 14 yaşındaki bir kız çoçuğu sunulmuştur.

Olgu: 14 yaşında cinsel olarak aktif olmayan bir kız çoçuğu acil servisimize ani başlayan vulvar ağrı ve lezyonlar ile başvurdu. Vulva ödem neden ile gergin ve orta derecede eritemli görünümde idi ve aftöz lezyonlar mevcuttu. HSV, gonore, klamidya, vulvanın aerobik kültürleri, sfiliz serolojisi ve immunologic testler negatifdi. Tüm kültürler, laboratuvar testleri, histopatolojik ve immünolojik incelemeler sonrası hastaya idyopatik vulvar aftozis tanısı konuldu.

Sonuç: Vulvar aftozis adolesanlardaki genital lezyonların nadir sebebidir. Genellikle kendini sınırlar fakat çocuklarda ve ailelerinde büyük stress nedenidir. Bu hastaların değerlendirilmesinde anamnez ve fizik muayene özenle yapılmalıdır. Tedavi yöntemi ağrının giderilmesi ve yara bakımındır.

Anahtar Kelimeler: Vulvar Aft; Genital Aft.

ABSTRACT

Introduction: A variety of viral infections have been blamed for vulvar aphtosis, such as Epstein-Barr virus, Cytomegalovirus, and influenza A. In this report we presented a 14-years old girl with the diagnosis of vulvar aphthosis.

Case: A 14-years old nonsexually active girl was admitted to our emergency unit with the complaint of sudden onset vulvar pain and lesions. On pelvic examination; vulvar area was tense with edema and moderately erythematous and vulvar aphtous lesions were present. HSV, gonorrhea, Chlamydia, and aerobic cultures of the vulva, syphilis serology and immunologic tests were negative. Since all cultures, laboratory tests, histopathological examinations were not diagnostic, the clinical diagnosis was idiopathic vulvar aphthosis.

Conclusion: Vulvar aphthosis is a rare cause of genital ulcers in adolescents. It is usually self limited but it causes great stress in children and their families. In these patients great care in taking the medical history and physical examination is crucial. Treatment consist of pain relief and wound care.

Key Words: Vulvar Aphthae; Genital Aphthous Ulcers.

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INTRODUCTION

Ulcus vulvae acutum (vulvar aphthosis, Lipschütz ulcer) is first described by Lipschütz in 1913 (1). These painful acute condition seen in young women is quite rare. In differential diagnosis; sexually and nonsexually transmitted infections and autoimmune casus should be investigated. Non-infectious vulvar ulcers can be associated with Behçet's disease or Crohn's diease (2). A variety of viral infections have been blamed, such as Epstein-Barr virus, Cytomegalovirus, and influenza A (1). In some cases these painful genital lesions are accompanied by systemic flu-like symptoms such as fever and malaise. In the presented case, a peripubertal nonsexually active child presented with extreme genital pain, vulvar ulcers with associated edema, and erythema of the labia minora. This entity is characterized by one or more vulvar ulcerations with spontaneous recovery (1).

CASE

A 14-years old girl admitted to our emergency unit with the complaint of sudden onset vulvar pain and aphtous lesions. Symptoms began just after her last menstruation. On pelvic examination, pudendum was edematous, and there were multiple aphtous lesions localized on inner side of labium minora. These white, aphtous lesions are 2-3 mm in diameter, and there were pinpoint petechia on their bases. The vulva was tense with edematous and moderately erythematous. No lymphadenopathy was noted. In her medical history there were no sexual contact, trauma, physical contact, trauma or sexually transmitted disease. Two days before first vulvar lesions appeared she had fatigue and mild fever. During her general examination another lesion was noticed on the right side of tongue. She was using dental braces. She had this lesion for a while without any oral aphtous lesion. The laboratory tests revealed a slightly elevated C-reactive protein whereas the complete blood count, blood chemistry profile, and urine analysis were normal. A biopsy was performed from one of the lesions and she was discharged with decongestant topical treatment. At day 5, she was admitted to our center again with vulvar pain and dysuria. Vulva was edematous and separation of the labia for inspection of the lesions revealed exquisitely tender, 1-2 cm ulcerations of the labium minora (Figure 1). The margins were well defined and flat; the center was 3 mm deep, grey, and covered with necrotic fibrous exudate. She was hospitalized, gonorrhea, chlamydia, HSV, and aerobic cultures were taken and topical antibiotic treatment was started. Urine analysis showed an urinary tract infection, oral antibiotic treatment was started. Betadine sitz baths followed by wet-to-dry debridement three times a day were initiated. She was consulted with dermatology and ophthalmology units. No retinitis or uveitis to support Behçet's syndrome was found and pathergy test was negative. HSV, gonorrhea, Chlamydia, and aerobic cultures of the vulva, plus syphilis serology, were negative. Histopathological examination showing chronic active inflammation and necrosis was not helpful for differential diagnosis. Since all cultures, laboratory tests, histopathological examinations were not diagnostic, the clinical diagnosis was vulvar aphthosis. At tenth day of treatment vulvar edema was diminished and improvement and epithelization of the ulcers was noticed. She was discharged from hospital with topical estrogen treatment. After three weeks the ulcers healed with minimal atrophic scars.

Figure 1–Labial ulcers at day 5.

DISCUSSION

Benjamin Lipschütz (1878-1931), Austrian dermatologist and bacteriologist, described clinical entity of acute genital ulcer occurring in an adolescent girl with a non-venereal infectious etiology in 1913 (1). Vulvar ulcers are not frequent in adolescents. Sexual

contact or abuse must be remembered when an adolescent presents with vulvar ulcers. A detailed clinical history must be obtained including description of vulvar symptoms and general symptoms, a confidential assessment of sexual experience, age at menarche, family history, and management prior to presentation. Oropharynx, lymph nodes, skin, joints, Tanner staging of breasts and pubic hair, and external genitalia must be evaluated. Clinical laboratory studies including complete blood count, C-reactive protein, urine analysis, serologic testing for syphilis, IgM and IgG antibodies to EBV viral capsid antigen, and EBV nuclear antigen, as well as IgM and IgG antibodies to Cytomegalovirus (CMV). Cultures from ulcers must be obtained before antibiotic treatment. Genital ulcerations similar to acute vulvar ulcers have been found in HIV-positive women so screening for HBV, HCV and HIVinfections must be performed.

Lipschütz disease has a characteristic hyperacute onset usually with fever and malaise. This disease usually affects adolescent girls. The ulcerations are very painful, with thick adherent slough and a surrounding red areola. They are most often solitary, on one side of the vulva, usually the inner aspect of the labium minus (3). Our case was first seen just after the primary lesions appeared. These painfull, white, aphtous lesions were 2-3 mm in diameter, and there were pin point petechia on their bases. There were no ulcer formation. There were erythema around the lesions and vulva was edematous. Secondary lesions were ulcerated and covered with necrotic fibrinous exudate at day 5 (Figure 1).

In the etiology of Lipschütz ulcers Ebstein-Barr virus is one of the most common agents blamed. There are many case reports about EBV. Common points in these cases were presence of lymphadenopathy (mostly generalized), fever and systemic symptoms, atypical lymphocytes, and positive serology (4). Resolution time of ulcers usually occurs in 2-3 weeks (4). Halvorsen et al. summarized 26 cases of EBV associated genital ulcers in females in 2006. In this reewv most of the patients had more than one ulcer and accompanying urinary symptoms (4). Other infections blamed in the etiology of the disease are Cytomegalovirus, and influenza A (5). There is also one study reporting the relationship of genital ulcers with Salmonella typhi (6).

The ulcers usually last for 2 to 3 weeks, and the severe pain presents for the first 10 to 14 days. Lesions are self limited and treatment with analgesics, topical xylocaine jelly, and regular tub soaks is usually enough . Initially ice packs are soothing and may reduce swelling. Despite the look of cellulitis, antibiotics are not necessary in most patients. It remains controversial whether antivirals or corticosteroids will prove helpful, because current reports of their use are anecdotal and case based (6).

In this case presence of an oral lesion on her tongue directed us to the diagnosis of Behçet's disease but in her medical history s there were no recurrent oral aphts (any shape, size or number at least 3 times in any 12 months) skin lesions, and eye inflammation (7). Pathergy reaction was also negative.

In conclusion, although vulvar aphthosisis a rare cause of genital ulcers in adolescents it must be kept in mind in patients with such lesions. It is usually self limited but it causes great stress in children and their families (8). In these patients great care during taking medical history and physical examination is crucial.

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