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Non-sexually related acute genital ulcers in a pubertal girl

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CASE STUDY

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ABSTRACT

Acute genital ulcers (AGU), also known as acute vulvar ulcers or Lipschütz ulcers, is an uncommon, non-sexually transmitted condition characterized by the sudden onset of painful necrotic ulcerations of the vulva or lower vagina. Their aetiology and pathogenesis are still unknown. They may be preceded by influenza-like symptoms, such as fever, headache or respiratory symptoms. We presented a case of pubertal girl with multiple painful ulcers on the vulva who visited our hospital. Other causes of vulvar ulcers were excluded by history taking, laboratory tests, and skin biopsy. We presented a case report of pubertal girl with non-sexually related AGU.

Key Words
Acute genital ulcers, non-sexual, pubertal girl

Implications for Practice:

1. What is known about this subject?
Lipschütz ulcers are ulcerations of vulva that occur without a clear source of infection. This is painful and there is no definite preventer or treatment.

2. What new information is offered in this case study?
To our knowledge, this is the first study on acute genital ulcers that occurred within a local area in Korea.

3. What are the implications for research, policy, or practice?
This study provides a clinical study of acute genital ulcers based on clinical, histopathologic and direct cytotoxic microbiologic findings.

Background

Vulvar ulcers are uncommon in females who are sexually inactive. When a young female presents with ulcers on the vulva, parents and clinicians are required to suspect a sexually transmitted infection due to sexual contact or abuse.[1] In 1913, Lipschütz reported a series of young, virginal women who developed vulvar ulcers without a clear infectious source.[2] Little progress has been made toward understanding these ulcers after the initial report. Thus, the natural history, aetiology, effective treatment and prevention measures remain incompletely understood. Today, the term “Lipschütz ulcer” refers to an ulceration of the vulva or lower vagina of non-venereal origin and is also known as “acute genital ulcer” (AGU).[3] AGU also includes vulvar ulcers that do not have an identifiable aetiology, based on clinical, histopathologic and direct cytotoxic microbiologic findings. AGU is very painful and distressing to women, and perplexing to the providers who care for them because of the rarity of these lesions. Non-sexually related AGU in a pubertal girl has not been reported previously in Korea. Therefore, we presented a case report of AGU in a 13-year-old woman.

Case details

A 13-year-old previously healthy girl, with no personal history of recurrent oral or genital aphthous lesions, presented with sudden onset of two painful vulvar ulcers. It was preceded by weakness, asthenia, sore throat with fever (>38°C), vomiting and diarrhoea for four days. She denied previous sexual contact or sexual abuse. Physical examination revealed two ulcerated necrotic lesions with

purulent exudate on the inner aspect of both labia minora with labial swelling, indurated, well demarcated irregular margins and approximately >10mm in diameter (Figure 1-A,B). Both ulcers were severely tender to palpation. Lymphadenopathy was not reported. Her hymen was intact and no vaginal discharge was noticed. Other skin or mucous membrane lesions were absent and ophthalmologic examination was negative. The findings of endoscopic gastro-duodenoscopy (EGD) and colonoscopy were negative for Crohn's disease. Behcet's disease was ruled out by rheumatologist based on negative result for antinuclear antibodies (ANA). Histopathologic examination from an ulcer edge was nonspecific inflammation with reactive squamous epithelial hyperplasia (Figure 2).

Analytical test results indicated an elevated C-reactive protein (8.28mg/dL) and leukocytosis (10,620cells/µL). After treatment started, C-reactive protein dropped to 1.71mg/dL. Liver enzymes were not altered. Herpes simplex virus (HSV), cytomegalovirus (CMV), Treponema pallidum (TP), Chlamydia trachomatis, Epstein-Barr virus (EBV), Mycoplasma hominis and Mycoplasma genitalium Polymerase chain reaction (PCR) studies were all negative. And human immunodeficiency virus (HIV) and syphilis serologic studies were all negative.

Treatments during hospitalization included supportive local care with sitz baths and urinary catheter insertion, pain control with oral and topical analgesics (Acetaminophen), anti-inflammatory drugs and prophylactic broad-spectrum antibiotics (Cefazedone). The patient showed a good clinical progress with improvement of the blood test results. She was discharged from hospital in good condition without pain after a total hospital stay of 8 days (Figure 1-C). The 3 week follow-up showed total resolution and re-epithelization of the vulvar lesion. No recurrences occurred during the following 6 months.

Discussion

In 1913, Lipschütz first described an acute disease comprising fever, genital ulceration and lymphadenopathy in an adolescent girl.[1] Aetiology and pathogenesis are still unknown, but most of acute vulvar ulcers are associated with infectious diseases such as Mycoplasma infection, paratyphoid fever, influenza A infection, and mostly with Epstein-Barr virus (EBV) infection.[4-11] AGU is characterized by an acute onset of flu-like symptoms such as malaise, fever, asthenia, myalgia, pharyngotonsillitis, lymphadenopathy and headache, with single or multiple, shallow and painful ulcers with raised, sharply demarcated borders.[12] Typically, ulcers occur on the medial aspects of the labia minora, and kissing ulcers on opposing surfaces like our case are common.[3,13] They usually occur in young women, predominantly virgins.[1,3] The histologic examination is not of diagnostic value due to nonspecific findings including superficial oedema and dilated capillaries with neutrophilic infiltration and ulceration.

Initial workup may include complete blood cell count; bacterial culture; serologic test for syphilis and HIV; PCR assays for HSV, EBV and TP; and skin biopsy from an ulcer edge if necessary. Although, we do not recommend all genital ulcers for biopsy, we recommend a biopsy if the case is ulcer that does not have sexual experience as in this patient.

Diagnosis of AGU is mainly clinical, after exclusion of other causes of vulvar ulcers. The differential diagnosis is extensive and includes venereal and non-venereal infections, noninfectious diseases, traumatic causes and malignant tumors (Table 1).

The treatment is mainly symptomatic. Topical antibiotics can be used, as well as oral corticosteroids and oral antibiotics for particularly painful AGU. The condition is self-limited and healing usually occurs spontaneously in 2 weeks.[12]

Conclusion

This study has some limitations. There is no photo left after treatment, causal inference not possible and generalization not passible. And pathologic, histologic, and immunological inaccurate precision mechanisms have yet to be identified. But there is a case report of a young virgin woman with a vulvar ulcer even without sexual contact. This study has educational value about rare diseases and it will provide basic data for the prevention and treatment of acute genital ulcers.

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PEER REVIEW

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CONFLICTS OF INTEREST

The authors have no conflict of interest to declare.

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None

PATIENT CONSENT

The authors, Yun Dan Kang, declare that:

1. They have obtained written, informed consent for the publication of the details relating to the patient(s) in this report.
2. All possible steps have been taken to safeguard the identity of the patient(s).
3. This submission is compliant with the requirements of local research ethics committees.

Table 1: Differential diagnosis of AGU in adolescence

Infection	
Sexually transmitted	Herpes simplex virus
	Syphilis
	Lymphogranuloma venereum
	Chancroid
	HIV
Non-sexually transmitted	Epstein-Barr virus
	Cytomegalovirus
	Influenza A
	Paratyphoid
Systemic disease	Crohn's disease
	Cyclic neutropenia
	PFAPA syndrome
	MAGIC syndrome
	Iron, folate, vitamin B12 deficiency
	Behcet's disease
	Pemphigus and pemphigoid
	Complex and simple aphthosis
Hormone-related	Autoimmune progesterone dermatitis
	Oestrogen hypersensitivity
Drug reaction	Non-steroidal anti-inflammatory drugs
	Contact or irritant dermatitis
Malignancy	Lymphoma / leukaemia
Trauma	Foreign body
	Sexual injury
	Caustic burns

PFAPA- periodic fever adenitis pharyngitis aphthous ulcer
MAGIC- mouth and genital ulcers with inflamed cartilage

Figure 1: Vulvar oedema (A), "Kissing" ulcers on medical aspects of labia minora (B) and Gross appearance after 8 days (C)

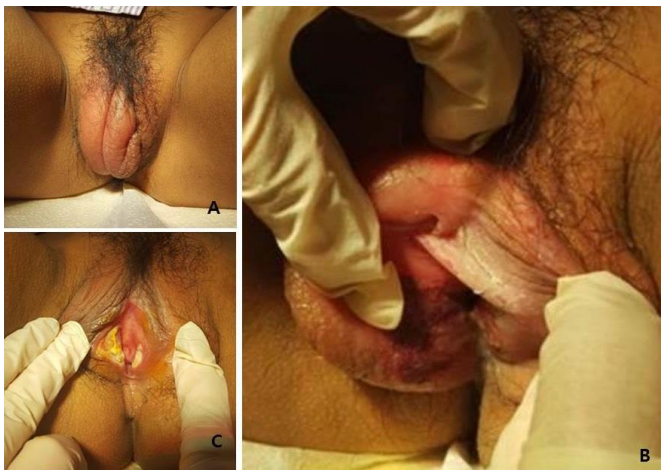

Figure 2: Non-specific inflammation with dense neutrophilic and lymphocytic infiltration (H&E: x100)