



## CASE REPORT

# Lipschütz Ulcer and Epstein-Barr Virus Infection: a Case Report

*Úlcera de Lipschütz e infecção pelo vírus Epstein-Barr: um relato de caso*

**Iris Santos Silva<sup>1</sup>**

[orcid.org/0000-0002-8138-8056](https://orcid.org/0000-0002-8138-8056)  
[iris.silva@ulsguarda.min-saude.pt](mailto:iris.silva@ulsguarda.min-saude.pt)

**João Virtuoso<sup>1</sup>**

[orcid.org/0000-0001-7720-4544](https://orcid.org/0000-0001-7720-4544)  
[joao.virtuoso@ulsguarda.min-saude.pt](mailto:joao.virtuoso@ulsguarda.min-saude.pt)

**Joana Filipe Ribeiro<sup>1</sup>**

[orcid.org/0000-0002-4651-9292](https://orcid.org/0000-0002-4651-9292)  
[joanafiliperibeiro@gmail.com](mailto:joanafiliperibeiro@gmail.com)

**Glória Silva<sup>1</sup>**

[orcid.org/0000-0002-0293-3141](https://orcid.org/0000-0002-0293-3141)  
[gloria.silva@ulsguarda.min-saude.pt](mailto:gloria.silva@ulsguarda.min-saude.pt)

**Rita S. Oliveira<sup>1</sup>**

[orcid.org/0000-0002-6758-8174](https://orcid.org/0000-0002-6758-8174)  
[rita.oliveira@ulsguarda.min-saude.pt](mailto:rita.oliveira@ulsguarda.min-saude.pt)

**Received on:** Feb. 7<sup>th</sup>, 2021

**Approved on:** Nov. 3<sup>rd</sup>, 2021

**Published on:** Dec. 20<sup>th</sup>, 2021.

## Abstract

**Aims:** Lipschütz ulcer (LU), also known as acute vulvar ulcer, is a rare cause of vulvar ulcerations of nonvenereal origin. Our aim is to alert about this manifestation of the disease and to prevent unnecessary treatment.

**Case description:** we present a 15 years old female, without relevant family and past history, admitted in the emergency room with a painful vulvar ulcer, preceded by five days of fever and sore throat. On physical examination, she had enlarged, and erythematous tonsils and bilateral anterior cervical lymphadenopathy and the genital examination revealed vulvar oedema and a deep ulcer with necrotic plaques in labium minus. The exclusion of transmitted sexual disease led to a diagnosis of Lipschütz ulcer. She started symptomatic treatment, oral antibiotic and corticoid therapy. She was discharged from the hospital after 6 days of admission and returned to a consult one month later when it was observed an almost complete resolution of the lesions. No recurrences occurred until 3 months.

**Conclusion:** LU is a misdiagnosed pathology, probably because doctors, in general, are not familiarized with that, and since the diagnosis is made by exclusion. Infectious, such as Epstein-Barr Virus infections, are proposed etiologies.

**Keywords:** adolescence, lipschütz ulcer, epstein-barr virus.

## Resumo

**Objetivo:** a úlcera de Lipschütz, ou úlcera vulvar aguda, é uma causa rara de ulceração de origem não venérea. O nosso objetivo é alertar para essa manifestação da doença e prevenir tratamentos desnecessários.

**Descrição:** adolescente, sexo feminino, 15 anos, sem história pessoal ou antecedentes familiares de relevo. Recorreu ao Serviço de Urgência por febre e odinofagia, com cinco dias de evolução, associada a uma úlcera vulvar dolorosa, que surgiu no próprio dia. O exame clínico revelou amígdalas hipertrofiadas e eritematosas, linfadenopatia cervical anterior, e o exame ginecológico apresentou edema da vulva e uma úlcera profunda, com placas necróticas nos lábios menores. A exclusão de uma doença sexualmente transmissível levou ao diagnóstico de úlcera de Lipschütz. Ficou internada no Serviço de Pediatria e iniciou tratamento sintomático, antibioterapia por sobreinfecção da úlcera e corticoterapia tópica. Teve alta após seis dias de hospitalização e foi reavaliada em consulta um mês depois, com resolução completa do caso. Não apresentou recorrência da úlcera até aos três meses seguintes.

**Conclusão:** a úlcera de Lipschütz é uma doença subdiagnosticada, muito provavelmente porque os médicos não estão familiarizados com ela e por ser um diagnóstico de exclusão. A infecção pelo vírus Epstein-Barr é uma das etiologias propostas.

**Palavras-chave:** adolescência, úlcera de lipschütz, vírus epstein-barr.



Artigo está licenciado sob forma de uma licença  
[Creative Commons Atribuição 4.0 Internacional](https://creativecommons.org/licenses/by/4.0/).

<sup>1</sup> Hospital Sousa Martins, Unidade Local de Saúde da Guarda; Paediatric Department, Guarda, Portugal.

## Introduction

Lipschütz ulcer (LU), also known as acute vulvar ulcer, is a rare cause of vulvar ulcerations of non-venereal origin. It can affect adolescents without a previous history of sexual contact. (1,2) Infectious and idiopathic causes are proposed etiologies for LU, for instance, it can be associated with Epstein-Barr virus (EBV), Cytomegalovirus, Influenza and Toxoplasmosis, among others (3,4,5).

EBV infection is often asymptomatic, however, in some patients, it can manifest as infectious mononucleosis, associated with systemic symptoms: fever, pharyngotonsillitis, adenopathy, myalgia, headache, hepatosplenomegaly and rarely pneumonia, hematologic and cardiovascular complications. (1,6,7) Lipschütz ulcer is a rare complication of primary EBV infection (7), however EBV is one of the most common agents associated with Lipschütz ulcer. (8)

## Case Report

A 15 years old female, 57 kilograms, without relevant family past history and with history of Cytomegalovirus Infection two years before, was admitted in the emergency room with fever (tympanic temperature: 39°C) and sore throat and was medicated/treated with ibuprofen. After five days, she returned to the hospital due to genital oedema and a vulvar ulcer. She denied sexual contact, other genital, or urinary symptoms, including pruritus or vaginal discharge, local trauma, arthralgia or oral ulcers. She also denied similar previous episodes. On physical examination, she had enlarged and erythematous tonsils and bilateral anterior cervical lymphadenopathy, without other adenopathies, hepatic or splenomegaly. Genital examination revealed vulvar oedema, and a 1.5cm of diameter, deep ulcer with necrotic plaques in *labium minus* (**Figure 1**).



**Figure 1** – Lipschütz ulcer at the onset in an 15-year-old female.

Laboratory findings included: leukocytosis (neutrophils – 75%, lymphocytes – 16% and monocytes 9%), elevated levels of serum C-reactive protein (22.7 mg/dl) and normal liver enzyme levels. Serological tests for Toxoplasmosis, *Treponema pallidum*, Human Immunodeficiency Viruses (HIV-1 and -2), Herpesviruses (HHV-1, -2, -3, and -5), *Mycoplasma sp*, *Brucella sp*, Parvovirus and Cytomegalovirus IgM were negative. The EBV serology (performed on day five of disease) was positive for IgM against viral capsid antigen (CMIA) & ELFA and IgG negative.

Histopathologic examination wasn't done.

She was admitted at pediatric service with the diagnosis of Lipschütz ulcer and started oral antibiotic: Doxycycline, 100 mg twice a day, fourteen days, and Clarithromycin, 500 mg twice a day, seven days, due to clinical suspicious of bacterial reinfection. Additionally, initiated topic treatment for ulcer with methylprednisolone and healing cream. Since she started vulvar pain during urination and presented inflammatory signs, she also started doing lidocaine cream (as necessary) and systemic prednisolone (40 mg/day, seven days). We verified clinical improvement, with apyrexia and decreased vulvar

pain. She was discharged from the hospital after six days of admission and returned to a consult. One month later when it was observed an almost complete resolution of the lesions. A recurrence occurred three months later, with resolution in one month, only treated with analgesic and healing cream. Until now she had regular pediatric consults (every six months), and after one year she had no more recurrences.

## Discussion

"Ulcus vulvae acutum" or "Lipschütz Ulcer" is a self-limited, and non-sexually transmitted condition. It can occur in adults or infants, and the prevalence is unknown. However, Lipschutz ulcers are considered rare; according to Vieira-Baptista et al. study, which included 110 women with vulvar ulcers, 30% were diagnosed with LU. (9) A recent study revealed that 4.39% of the 273 patients included can be diagnosed with LU. (1).

The pathogenesis of acute genital ulceration is unclear, there are reports of genital ulceration associated with viruses (Epstein-Barr virus, Cytomegalovirus, Influenza, Paratyphoid fever, Toxoplasmosis, among others) and bacterial infection (10-12). Most reports associate acute genital with primary infection of EBV (1, 13). Three hypotheses have been suggested for the pathogenesis of LU due to EBV infection: the first one is a type-III hypersensitivity reaction, caused by the immune complexes produced during the acute phase of EBV infection; the second one says that LU can result of the combination between cytolysis due to EBV replication in vulvar keratinocytes and reactive inflammatory response to the expression of viral antigens; and, finally, the third one admits that LU might be a type of aphthosis. (6,7) Generally, ulcers associated with EBV have a diameter >1 cm, are often quite deep and necrotic, very painful and may cause urinary symptoms (for example dysuria and urine retention). Usually, they involve the *labia minora*, but can extend to the *labia majora*, perineum, vestibule, and lower vagina. They also can be preceded by general symptoms (fever, malaise, tonsillitis, lymphadenopathy) (4, 9, 14).

Majority of cases occurred in female patients younger than 20 years; this fact can be justified because viral infections mentioned above are more frequent in young people (1, 10).

This diagnosis is made by exclusion, so we cannot forget other causes of acute genital ulcer, such as Herpes simplex virus and aphthosis (9,10). A careful sexual history should also be taken, with the assurance of confidentiality, and syphilitic chancre, lymphogranuloma venereum, chancroid, hepatitis and HIV should be discarded (15). Although a detailed history and complete physical examination are the first and an important step for diagnosis, we also have to do laboratory investigations to exclude diverse causes of genital ulceration (Behçet syndrome, inflammatory bowel disease, autoimmune diseases, virus or bacterial infection).

We decided not to do histopathologic examination, since the biopsy is in general unhelpful because biopsies of acute vulvar ulcers are often nonspecific and include necrosis of the epithelium (4,10).

The condition is self-limited, and healing occurs spontaneously in two to six weeks. The treatment is mainly symptomatic, with healing cream and topical lidocaine. In our case, we also use antibiotic and corticosteroid therapy since she had a painful and necrotic ulcer. According to literature topical antibiotics can be used, as well as oral corticosteroids and antibiotics, for peculiarly painful acute genital ulcers (15, 16). These adolescents should keep follow-up in consultation for surveillance of the appearance of new genital ulcerations or diagnostic criteria for other diseases.

In conclusion, LU is a misdiagnosed pathology, probably because physicians are not familiarized with that and since the diagnosis is made by exclusion. This case aims to alert the possibility for this type of symptom, differential diagnosis, and prevent unnecessary treatment.

## Notes

This study was presented at the 9<sup>th</sup> National Congress of Adolescent Medicine, Portuguese Society of Adolescent Medicine of the Portuguese Society of Pediatrics (SPMA-SPP), Porto, Portugal, in May, 31<sup>th</sup> 2019.

## Funding

This study did not receive financial support from external sources.

## Conflicts of interest disclosure

The authors declare no competing interests relevant to the content of this study.

## Authors' contributions

All the authors declare to have made substantial contributions to the conception, or design, or acquisition, or analysis, or interpretation of data; and drafting the work or revising it critically for important intellectual content; and to approve the version to be published.

## Availability of data and responsibility for the results

All the authors declare to have had full access to the available data and they assume full responsibility for the integrity of these results.

## References

- Schindler Leal AA, Piccinato CA, Beck APA, Gomes MTV, Podgaec S. Acute genital ulcers: keep in mind. *Arch Gynecol Obstet.* 2018;298(5):927-31. <https://doi.org/10.1007/s00404-018-4866-6>
- Bounniyt H, Mael-Ainin M, Bouhallab J, Hassam B. Lipschütz ulcer: a rare case of genital ulcers. *Presse Med.* 2015;44(1):117-8. <https://doi.org/10.1016/j.lpm.2014.04.018>
- Burquete Archel E, Ruiz Goikoetxea M, Recari Elizalde E, Beristain Rementería X, Gómez Gómez L, Iceta Lizarraga A. Lipschütz ulcer in a 17-month-old girl: a rare manifestation of Epstein-Barr primoinfection. *Eur J Pediatr.* 2013;172(8):1121-23. <https://doi.org/10.1007/s00431-013-2013-8>
- Vismara SA, Lava SAG, Kottanattu L, Simonetti GD, Zraggen L, Clericetti CM, et al. Lipschütz's acute vulvar ulcer: a systematic review. *Eur J Pediatr.* 2020;179(10):1559-67. <https://doi.org/10.1007/s00431-020-03647-y>
- Truchuelo MT, Vano-Galván S, Alcántara J, Pérez B, Jaén P. Lipschütz ulcers in twin sisters. *Pediatr Dermatol.* 2012;29(3):370-2. <https://doi.org/10.1111/j.1525-1470.2011.01715.x>
- Sárdy M, Wollenberg A, Niedermeier A, Flaig MJ. Genital ulcers associated with Epstein-Barr virus infection (ulcus vulvae acutum). *Acta Derm Venereol.* 2011;91(1):55-9. <https://doi.org/10.2340/00015555-0979>
- Bolis V, Karadedos C, Chiotis I, Chaliasos N, Tsaouris S. Atypical manifestations of Epstein-Barr virus in children: a diagnostic challenge. *J Pediatr (Rio J).* 2016; 92(2):11-21. <https://doi.org/10.1016/j.jped.2015.06.007>
- Arellano J, Fuentes E, Moreno P, Corredoira Y. Úlceras de Lipschütz, un diagnóstico para considerar en la población pediátrica. *Arch Argent Pediatr.* 2019;117(3):e305-e308. <https://doi.org/10.5546/aap.2019.e305>
- Vieira-Baptista P, Lima-Silva J, Beires J, Martinez-de-Oliveira J. Lipschütz ulcers: should we rethink this? An analysis of 33 cases. *Eur J Obstet Gynecol Reprod Biol.* 2016; 198:149-52. <https://doi.org/10.1016/j.ejogrb.2015.07.016>
- Farhi D, Wendling J, Molinari E, Raynal J, Carcelain G, Morand P, et al. Non-sexually related acute genital ulcers in 13 pubertal girls: A clinical and microbiological study. *Arch Dermatol.* 2009;145(1):38-45. <https://doi.org/10.1001/archdermatol.2008.519>
- Vieira-Baptista P, Machado L, Costa AR, Beires J, Martinez-de-Oliveira J. Mycoplasma pneumoniae: a rare cause of vulvar ulcers or an undiagnosed one? *J Low Genit Tract Dis.* 2013;17(3):330-4. <https://doi.org/10.1097/lgt.0b013e3182710896>
- Limão S, Ventura A, Queirós G, Cunha F. Lipschütz ulcer and group A streptococcal tonsillitis. *BMJ Case Rep.* 2018;2018:bcr2017223579. <https://dx.doi.org/10.1136/bcr-2017-223579>
- Jerdan K, Aronson I, Hernandez C, et al. Genital ulcers associated with Epstein-Barr virus. *Cutis.* 2013; 91(6):273-6.
- Charamanta M, Soldatou A, Michala L. Vulvar Ulcers in Children: Dramatic But Self-Limited. *Pediatr Emerg Care.* 2021;37(2):70-2. <https://doi.org/10.1097/pec.0000000000002004>
- Hueto Najarro A, González García G, Breton Hernández P, et al. Úlceras genitales de Lipschütz: reporte de un caso pediátrico. *Arch Argent Pediatr.* 2017;115(6):e436-e439.
- Brinca A, Canelas MM, Carvalho MJ, Vieira R, Figueiredo A. Lipschütz ulcer (ulcus vulvae acutum): A rare cause of genital lesion. *An Bras Dermatol.* 2012; 87(4):622-4. <https://doi.org/10.1590/S0365-05962012000400018>

---

### Íris Santos Silva

Master in Medicine from Faculty of Medicine of the University of Porto (FMUP); pediatric resident in Hospital Sousa Martins (ULS), Guarda, Portugal.

---

### João Virtuoso

Master in Medicine from Faculty of Medicine of University of Lisboa; pediatric resident in Hospital Sousa Martins (ULS), Guarda, Portugal.

---

### Joana Filipe Ribeiro

Master in Medicine from Faculty of Medicine of the University of Porto (FMUP); pediatric resident in Hospital Sousa Martins (ULS), Guarda, Portugal.

---

### Glória Silva

Master in Medicine from Institute of biomedical Sciences Abel Salazar (ICBAS) of University of Porto; pediatrician in Hospital Sousa Martins (ULS), Guarda, Portugal.

---

### Rita S. Oliveira

Master in Medicine from Faculty of Medical Sciences of University NOVA de Lisboa; pediatrician in Hospital Sousa Martins (ULS), Guarda, Portugal.

---

### Mailing address

Íris Anabela Santos Silva

Av. Rainha Dona Amélia 19

6300-035

Guarda, Portugal

*Os textos deste artigo foram revisados pela Poá Comunicação e submetidos para validação do(s) autor(es) antes da publicação.*