# Lipschütz Ulcer associated with SARS-CoV-2 infection – a case report

# Úlcera de Lipschütz associada a infeção por SARS-CoV-2 – um caso clínico

Joana da Cunha Simões¹, Joana Rodrigues¹, Vera Mourinha¹, Eunice Capela¹, Amália Pacheco¹ Centro Hospitalar Universitário do Algarve

## **Abstract**

Lipschütz ulcer (LU) is a diagnosis of exclusion, characterized by the sudden appearence of deep, wide and painful vulvar ulcers associated with a non-sexually transmited infectious disease. We describe a case of a woman with fever, myalgias and odynophagia associated with SARS-CoV-2 infection and deep, painful vulvar ulcers in a kissing pattern. She did topic corticosteroids and lidocaine treatment and had a complete resolution. No other cause for the vulvar ulcers was found. We found four other similar cases in recent literature. LU is often underdiagnosed, and the aim of the article is to highlight the possibility of this diagnosis in a patient with COVID-19 after exclusion of other possible causes.

**Keywords:** Vulva; Ulcer; Vulvodynia; SARS-CoV-2; COVID-19; Case report.

#### Resumo

Úlcera de Lipschütz (UL) trata-se de um diagnóstico de exclusão, que se caracteriza pelo aparecimento súbito de úlceras vulvares dolorosas, profundas e de grandes dimensões, normalmente associadas a uma doença infeciosa de transmissão não sexual. Neste artigo descrevemos o caso de uma mulher com febre, mialgias e odinofagia associadas a SARS-CoV 2 e úlceras vulvares profundas, dolorosas, com padrão em espelho características de UL. Fez tratamento com corticoides tópicos e lidocaína com resolução completa. Não foi encontrada outra causa para as úlceras vulvares. Encontrámos na literatura recente quatro outros casos semelhantes. A UL é muitas vezes subdiagnosticada pelo que o objetivo do artigo é sublinhar a possibilidade deste diagnóstico numa paciente com COVID-19 após exclusão de outras causas possíveis.

Palavras-chave: Vulva; Úlcera; Vulvodinia; SARS-CoV-2; COVID-19; Caso-clínico.

# **INTRODUCTION**

Lipschütz ulcer (LU) was first described in 1912 by Benjamin Lipschütz. It was identified as a rare cause of non-venereal vulvar ulcerations that usually affect young individuals and adolescents with no prior sexual contact<sup>1</sup>. Today it is known to occur more often in ado-

lescent and young women but can affect individuals of any age group and sexual status<sup>2</sup>.

There is no consensus about the diagnosis criteria in the literature, and it is believed that this condition is underdiagnosed due to the lack of awareness among healthcare providers<sup>2,3</sup>.

LU is a non-sexually acquired disease, that manifests as sudden-onset, painful, acute vulvar ulcers, usually preceded by fever, malaise, and other unspecific symptoms

<sup>1.</sup> Centro Hospitalar Universitário do Algarve.

like asthenia, headache, myalgias and adenopathy. The ulcers are typically deep and necrotic, wider than 1 cm and appear in a mirror distribution ("kissing lesions"), primarily affecting the middle region of the labia minora. However, assymmetric or unilateral and non-necrotic ulcers have also been described.

Symptomatic treatment options, such as pain relief with acetaminophen or ibuprofen, topical treatment with 2% lidocaine gel or benzocaine, and oral or topical steroid use to reduce inflammation (such as Clobetasol 0,05% ointment or prednisolone 40 mg peros 10 days), are the main options available. The use of colchicine (1 mg peros), pentoxifiline (400 mg peros) or topical silver nitrate for symptoms relief has also been described. The condition is usually self-limited, and relapses are rare<sup>1,2</sup>.

The mechanism behind the manifestation of LU is still unclear. It is thought to be triggered by an exacerbated immune response to agents such as Epstein-Barr virus (EBV), cytomegalovirus, Mycoplasma pneumoniae and toxoplasma gondii, although it is thought it may be associated with other agents as influenza virus and adenovirus<sup>1,3</sup>.

It is a diagnosis of exclusion and sexually transmitted infection, Behçet's syndrome, Crohn's disease and other specific ulcerative diseases must be ruled out.

We found, in recent literature, four cases of genital ulcers associated with severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) infection. 4,5,6,7 In this report, we present a case of a woman who developed acute-onset, painful vulvar ulcers during concurrent SARS-CoV-2 infection.

# **CASE DESCRIPTION**

A 34 year old women resorted to emergency room complaining of vulvar pain. She was a non-smoker, with no personal or family history of autoimmune diseases or previous genital ulcer, and no regular use of medication. At the time she had no sexual partner and reported not having sexual intercourse several months prior.

The woman presented with a fever, myalgia and odynophagia and was diagnosed with COVID-19 two days prior of presenting with vulvar pain.

At the observation the patient had deep, bilateral symmetric painful ulcers with a central necrotic area



**FIGURE 1.** Vulvar lesions at first examination. We can see symmetric ulcers in the labia minora, with a central necrotic area.

(Figure 1). There were no cervical or vaginal abnormalities, and no ulcers in any other location, including the oral or anal area, and no lymphadenopathies were noted. There was no pathologic discharge.

Blood tests showed mild leukocytosis ( $11800 \times 109/L$ ) and neutrophilia ( $8600 \times 109/L$ ) and C-reactive protein of 41 mg/L.

She had negative serologic tests for acute infection for Herpes simplex virus (positive immunoglobulin G and negative M (IgG+/IgM-), Treponema pallidum (IgG-/IgM), EBV (IgG+/IgM-) and HIV.

There were no clinic criteria of Behçet's disease or symptoms related to Crohn's disease.

Overall, the patient's clinical presentation, characteristic lesions, negative serologic tests for common infectious causes of genital ulcers, and lack of other signs and symptoms suggestive of underlying autoimmune or inflammatory conditions, led us to conclude that Lipschütz's ulcers related to COVID-19 was the most probable diagnosis.

Considering the patient's complaints of pain, we prescribed topical corticosteroid treatment (hydrocortisone 10 mg/g) and lidocaine (gel, 20 mg/g). The topical corticosteroids were administered twice a day, while the lidocaine was used as needed for pain relief. The patient showed significant improvement of the lesions after one week of treatment (Figure 2), with no



**FIGURE 2.** Examination after three weeks. We can see complete remission, without scaring.

vulvar pain and complete remission of systemic symptoms associated with COVID-19. The treatment was continued for three weeks until there was complete remission of the ulcers.

#### **DISCUSSION**

As described before, there are a few known infecions associated with Lipschütz ulcer and until recently CO-VID-19 was not one of them. The case report presented is of most importance as it highlights a rare manifestation of COVID-19 that can be easily misdiagnosed, with only four cases reported in the literature, so far. Lipschütz ulcer is a rare cause of vulvar ulcerations, mostly affecting young people and adolescents without prior sexual contact. However, it can manifest in patients of any age group and sexual status.

The presentation of acute vulvar ulcers, associated with fever and malaise, in the absence of sexually transmitted infections and other specific ulcerative diseases, is a hallmark of Lipschütz ulcer.

It is noteworthy that the diagnosis of Lipschütz ulcer is still challenging, and therefore, it is often underdiagnosed. Clinicians must be aware of this pathology, particularly when examining young patients with vulvar pain, and take the necessary steps to exclude other diagnoses.

#### **REFERENCES**

- 1. Vismara SA, Lava SAG, Kottanattu L, Simonetti GD, Zgraggen L, Clericetti CM, Bianchetti MG, Milani GP. Lipschütz's acute vulvar ulcer: a systematic review. Eur J Pediatr. 2020 Oct;179(10):1559-1567. doi: 10.1007/s00431-020-03647-y. Epub 2020 Apr 15. PMID: 32296983.
- 2. Vieira-Baptista P, Lima-Silva J, Beires J, Martinez-de-Oliveira J (2016) Lipschütz ulcers: should we rethink this? An analysis of 33 cases. Eur J Obstet Gynecol Reprod Biol 198:149-152
- 3. Schindler Leal AA, Piccinato CA, Beck APA, Gomes MTV, Podgaec S. Acute genital ulcers: keep Lipschütz ulcer in mind. Arch Gynecol Obstet. 2018 Nov;298(5):927-931. doi: 10.1007/s00404-018-4866-6. Epub 2018 Aug 24. PMID: 30143859.
- 4. Krapf, Jill M et al. "Reactive non-sexually related acute genital ulcers associated with COVID-19." BMJ case reports vol. 14,5 e242653. 5 May. 2021, doi:10.1136/bcr-2021-242653
- 5. Falkenhain-Lópes D, Agud-Dios M, Ortiz-Romero PL, Sánchez-Velázquez A. COVID-19-related acute genital ulcers. J Eur Acad Dermatol Venereol. 2020;34(11):e655-6. 4.
- 6. Christl J, Alaniz VI, Appiah L, Buyers E, Scott S, Huguelet PS. Vulvar Aphthous Ulcer in an Adolescent With COVID-19. J Pediatr Adolesc Gynecol. 2021:S1083-3188(21)00122-4
- 7. Jacyntho CM, Lacerda MI, Carvalho MS, Ramos MR, Vieira-Baptista P, Bandeira SH. COVID-19 related acute genital ulcer: a case report. einstein (São Paulo). 2021;19:eRC6541.

#### **AUTHORS CONTRIBUTION**

JS: Draft of the case description VM, EC: Critical review of the paper

#### **DATA CONFIDENTIALITY**

The authors declare having followed the protocols in use at their working center regarding patients' data publication.

#### **INFORMED CONSENT**

Obtained.

# **COMPETING INTERESTS**

The authors declare that there are no competing interests.

#### **FUNDING SOURCES**

The authors declare that there were no external sources of study for the performance of this article.

#### **CORRESPONDENCE TO:**

Joana da Cunha Simões E-mail: joana.c.s@netcabo.pt https://orcid.org/0000-0001-9920-115X

**RECEIVED:** 11/01/2023 **ACCEPTED:** 15/04/2023