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Lipschütz ulcers: should we rethink this? An analysis of 33 cases

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ABSTRACT

Objectives: To describe the presentation and characteristics of patients with Lipschütz ulcers (LU) and to evaluate the role of a standard protocol in the aetiological diagnosis.
Study design: Retrospective analysis of all cases of diagnosis of LU at our Vulvar Clinic during a five-year period.
Results: Of 110 women with vulvar ulcers, 33 (30.0%) had a diagnosis of LU. The mean age was 29.1 ± 15.14 years (10–79 years). Nine (27.3%) were 35 years old or more. The majority had had their sexual debut (28, 84.8%). Ten patients (30.3%) referred had at least one previous similar episode. Twenty-five (75.7%) had non-gynecological symptoms in the week before. The ulcers were located most frequently on the vestibule (19, 57.6%) and the labia *minora* (10, 30.3%). Isolated lesions on the left side were uncommon (3, 9.1%). Most had multiple (22, 66.7%) lesions. The mean time to full healing of the lesions was 15.6 ± 6.20 days.

A microbiological possible cause was identified in 9 (27.3%) patients: CMV (3 cases), *Mycoplasma pneumoniae* (3 cases), EBV (2 cases) and PVB19 (1 case). The protocol did not include systematic biopsies, blood count differentials, C-reactive protein and liver enzymes, which may have lead to some missed diagnosis.
Conclusions: LU can be found in women of any age, most of them sexually active. In most cases the lesions are preceded by non-gynecological symptoms and recurrence is common. Most lesions occur on the vestibule and labia *minora*, being rarely found isolated on the left side. Viruses seem to be the most frequent associated agents but *Mycoplasma pneumoniae* serology should be considered. In one case there was a possible role for PVB19. A standard protocol can effectively exclude sexually transmitted diseases and lead to a diagnosis in up to a third of cases.

Introduction

Lipschütz ulcers (LU) (also known as ulcus vulvae acutum, acute vulvar ulcers, primary aphthous ulcers or reactive non-sexually related acute genital ulcers) (Figs. 1 and 2) were first described in 1913 [1]. These ulcers are considered to have a sudden onset, to be acute, painful and nonsexually transmitted. Systemic symptoms, such as fever, myalgias, headache, diarrhea, oral aphthae, enlarged lymph nodes, tonsillitis or respiratory symptoms can be present. Healing occurs spontaneously, sometimes with scarring; the chance of recurrence is questionable. It is considered to be more frequent in girls or young women, who frequently are virgins. The diagnosis is one of exclusion, after ruling out sexually transmitted infections, idiopathic aphthosis, Behçet's syndrome, extra-genital Crohn's disease[2] or any other specific diagnosis.

LU are frequently undiagnosed and thus its true incidence is unknown. However, it can be a great cause of anxiety to patients and their parents (when in children) as well as to doctors, to whom it is sometimes an unknown entity [3].

In recent years, these ulcers have been related to infectious agents, including Epstein–Barr virus (EBV) [4], *Mycoplasma pneumoniae* [5] cytomegalovirus (CMV) [6] and *Salmonella* [7], among others. The mechanisms that lead to the formation of ulcers distant to the site of the primary infection are poorly understood.

It is useful to relate these lesions to a non-sexually transmitted agent, as it can help to relief some anxiety. Knowing that there is a specific agent related to the lesions can be helpful also to avoid an over-diagnosis of Behçet's syndrome, if other characteristics of the latter are also present.

There are no large series of cases of LU published. Most of the data derives from sporadic or anecdotal cases, and a significant number of those come from the pediatric literature.

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Fig. 1. Unilateral, single Lipschütz ulcer.

Fig. 2. Bilateral Lipschütz ulcers.

The authors intend to describe the characteristics of the affected patients, the form of presentation of the condition and to evaluate up to what point a standard protocol is useful in terms of reaching an aetiological diagnosis.

Materials and methods

Retrospective analysis of all cases with "vulvar ulcer" or "vulvar erosions" (past or present), diagnosed at the Vulvar Clinic of our institution during a five-year period (March 2009 to March 2014) and selection of those with a final diagnosis of "Lipschütz ulcer".

The minimum study protocol of these patients included: serology for human immunodeficiency virus (HIV), herpes simplex virus (HSV) 1 and 2, parvovirus B19 (PVB19), cytomegalovirus (CMV), Epstein–Barr virus (EBV), *Toxoplasma gondii* and *Mycoplasma pneumoniae*. For all serology tests, in case the interpretation of the results was doubtful, it was repeated in 2–4 weeks

A treponemal (TPHA) and/or a non-treponemal (VDRL) test was mandatory (if negative and high suspicion, it was repeated in 2 weeks), as well as a PCR sample from the lesion to test for the presence of HSV. Cultures, PCR testing for *Treponema pallidum*, darkfield microscopy and AgHBs were performed in some cases, but not considered mandatory. No biopsies were taken, unless there was suspicion of vulvar intraepithelial neoplasia or cancer.

Statistical analysis was performed using Microsoft® Excel® 2011 (Microsoft Corporation©, Redmond, Washington) and IBM® SPSS® 20.0 (IBM Corporation©, Armonk, NY). The Fisher's exact test was used for categorical variables. A p value <0.05 was considered statistically significant.

Results

During the evaluated time period, 110 patients were referred for consultation with a diagnosis of past or present vulvar ulcers or erosions. Of these 33 (30.0%) had a final diagnosis of Lipschütz ulcer, 34 (30.9%) of genital herpes, 32 (29 1%) of Behçet's syndrome/bipolar aphthosis, two (1.8%) of neuropathic ulcers, one (0.9%) that was considered to be drug related (fixed drug eruption to ceftriaxone) and one (0.9%) that was a long term CMV ulcer. In seven cases a diagnosis could not be attained. Most of the patients with the diagnosis of LU were referred from the Emergency Room (29, 85.3%). The mean age of these patients was 29.1 ± 15.14 years, ranging from 10 to 79 years. Almost a quarter of the patients were aged 20–24 years old and approximately another quarter were younger. Nine (27.3%) patients were 35 years old or more (Table 1). The majority of patients were or had been sexually active (28, 84.8%); the five that had never had intercourse were all

Table 1
Characterization of the population.

	n (*N* = 33)	%
Age		
10–14 years	5	15.2
15–19 years	3	9.1
20–24 years	8	24.2
25–29 years	4	12.1
30–34 years	4	12.1
35–39 years	3	9.1
40–44 years	2	6.1
>44 years	4	12.1
Sexually active (ever)	28	84.8
Parous	13	39.3
Autoimmune conditions/diseases	6	18.2
Smokers	7	21.2