Chikungunya associated vulvitis: case report and literature review

Vulvite associada a Chikungunya: relato de caso e revisão de literatura

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ABSTRACT

Introduction: Chikungunya virus is spreading worldwide due to migration and globalization and could be presented with systemic and with unusual symptoms. **Objective:** To report a case of virus-transmitted infection detected in a woman during the gynecological examination at a vulvar clinic. **Case report:** A 73-year-old Caucasian woman attended a vulvar clinic because of dyspareunia and vulvar burning. Ulcers were observed on labia minora and perineum. A Chikungunya was diagnosed by seroconversion in paired specimens. She was prescribed prednisolone 40 mg once a day for 10 days. After oral steroid treatment, the woman had no body rashes or lesions on her genitals. **Conclusion:** This study emphasized that rare signs of unusual vulvitis with ulcers could be associated with Chikungunya infection.

Keywords: Chikungunya virus; vulvitis; skin ulcer; alphavirus; virus diseases.

RESUMO

Introdução: O vírus Chikungunya está se espalhando pelo mundo por conta da migração e da globalização, podendo apresentar sintomas sistêmicos e incomuns. **Objetivo:** Relatar um caso de infecção pelo vírus detectado em uma mulher por ocasião do exame ginecológico em clínica de patologia vulvar. **Relato do caso:** Uma mulher caucasiana de 73 anos foi a uma clínica vulvar por causa de dispareunia e queimação vulvar. Úlceras foram observadas nos pequenos lábios e no períneo. O diagnóstico de Chikungunya foi realizado por soroconversão em espécimes pareados. Foi prescrita prednisolona 40 mg uma vez ao dia por dez dias. Após o tratamento com esteróides orais, a mulher não apresentou erupções ou lesões nos órgãos genitais. **Conclusão:** Este estudo enfatizou que quadros raros de vulvite com úlcera podem estar associados à infecção por Chikungunya.

Palavras-chave: vírus Chikungunya; vulvite; úlcera cutânea; alfavírus; viroses.

INTRODUCTION

Chikungunya virus is an arbovirus of the Togaviridae family, transmitted by the *Aedes Aegypti* mosquito[1]. The virus was first detected in Tanzania in 1952[2]. However, it has spread to the Americas, Asia, and Europe due to tourism and globalization[2].

The Chikungunya virus usually manifests itself as a systemic disease. Infected people develop fever, polyarthralgia, myalgia, and maculopapular rash on the skin. Despite mucocutaneous damage, genital lesions are significantly rare (approximately 2.7%)[3], mainly described in men. The objective is to describe vulvar lesions related to Chikungunya viral infection.

CASE HISTORY

A 73-year-old white woman attended a vulvar specialty clinic at *Hospital Universitário Antonio Pedro*, Niterói, Rio de Janeiro, Brazil, (August 3rd, 2018) complaining about dyspareunia, vulvar burning, and vulvar spotting that had started two weeks before the visit. One month prior to her appointment, she presented a low fever that lasted for one week. After the fever subsided, she developed maculopapular rash with pruritus on her limbs and torso, polyarthralgia, myalgia, asthenia, nausea, and abdominal pain. After one week, nausea and abdominal pain ceased, but the other symptoms persisted and she also presented dyspareunia, vulvar burning, and vulvar spotting (**Figure 1**).

Due to persistent pain in the vulva, she self-medicated with nimesulide 100 mg every 12 hours for three days and ibuprofen 400 mg once a day for five days with no positive response. She had no other comorbidities, except hypothyroidism (treated with L-thyroxine 50 mcg/d), and had been using hormone replacement therapy (HRT) since the age of 54 (estradiol 1 mg and norethindrone acetate 0.5 mg, continuously).

Vaginal clinical examination revealed good trophism, without petechiae or erosions. The vulva presented hyperemia, edema, and spotting from ulcers in the labia minora and perineum. The size of the ulcers varied from 0.8 cm in diameter, their shape was oval, and there were three of them (**Figure 1**).

Maculopapular rashes were observed on the neck, torso, and extremities (**Figure 1**). The culture from vulva and vagina was negative for *Candida* spp. and group A *Streptococcus*. The vaginal wet mount smear was normal, revealing *Lactobacillus spp.* predominance and the absence of leucocytes, *Candida* spp., and *T. vaginalis*.

The complete blood count on August 2nd, 2018, was in the normal range: erythrocytes $4.34x10^9$/L; hemoglobin 133 g/L; hematocrit 39.6%; thrombocytes $247x10^{12}$/L; leukocytes 5.8x10/L. The creatinine (0.68 mg/dl), alanine transaminase (9 U/L), and aspartate aminotransferase (10 U/L) were also in the normal range.

The patient's blood test was negative for *Herpes simplex* 1, 2 (HSV), HIV-1/2, and syphilis. Antibodies tests performed on July 25th, 2018 were nonreactive for Chikungunya virus IgM and IgG, as well as for Dengue and Zika viruses IgM, with a reactive test

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result for Dengue and Zika viruses IgG. Antibodies tests against Chikungunya, Dengue, and Zika viruses were repeated on August 23rd, 2018, rendering a reactive result for Chikungunya virus IgM and IgG, and no change from the prior results against Dengue and Zika viruses. A Chikungunya diagnosis was established by seroconversion in paired specimens (**Table 1**).

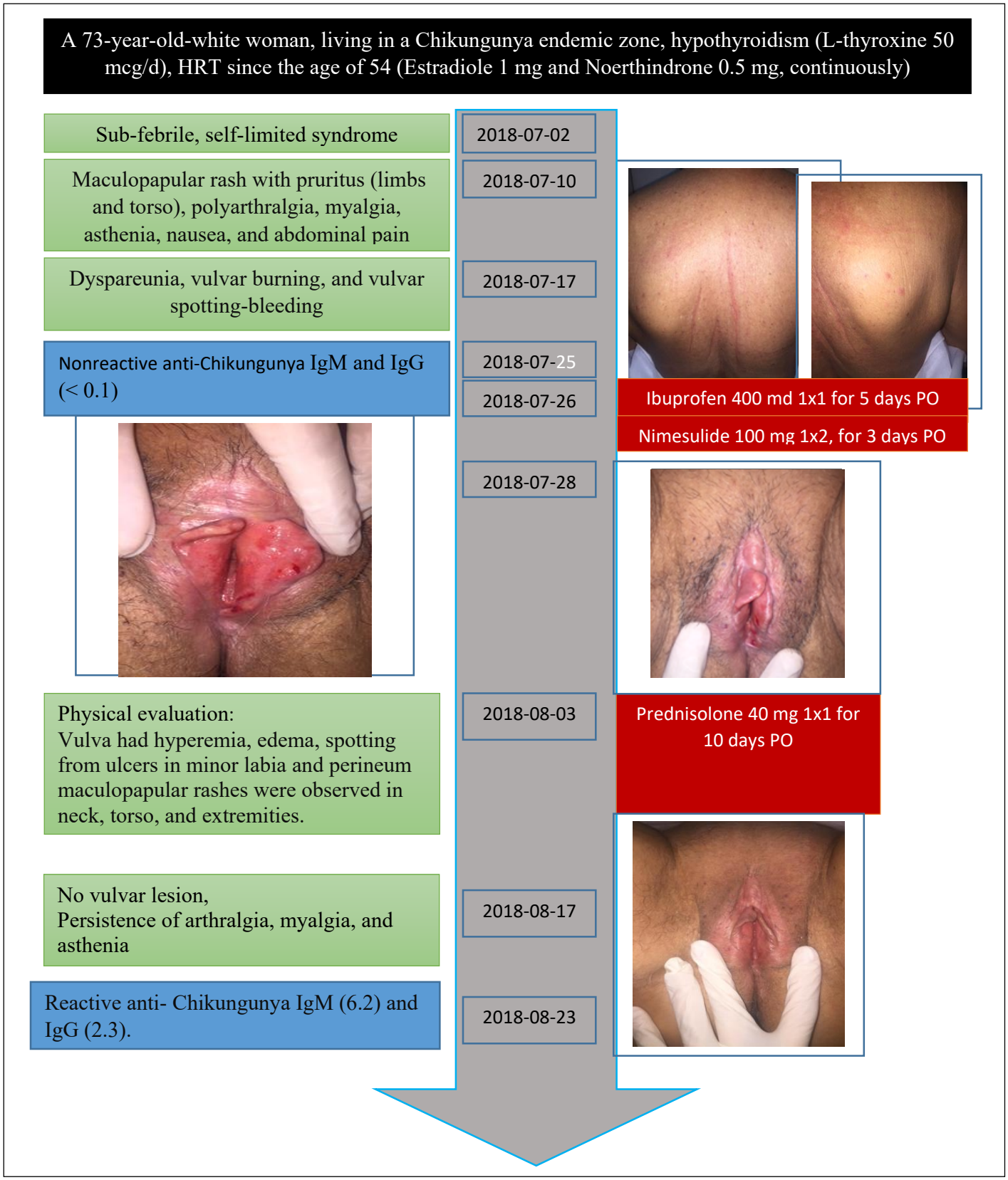

Figure 1 – Timeline of the patient's case.

The patient had used non-steroid anti-inflammatory drugs for one month before coming to the clinic, although she had no improvement in the vulvar ulcers and had developed inflammatory arthritis. She was prescribed prednisolone 40 mg once a day for 10 days. When she returned for a control visit two weeks after starting the oral steroid, she had no body rashes or lesions on the genitals (**Figure 1**), but still maintained arthralgia, myalgia, and asthenia.

DISCUSSION

This work presented a case report regarding genital lesions in a citizen from the city of Rio de Janeiro, Brazil, which is an endemic zone for the Chikungunya virus. This case demonstrates the need for gynecologists to be aware of mosquito-borne viral infections that may cause vulvar symptoms. The differential diagnosis of vulvar ulcers must also include the possibility of Chikungunya, Zika, and Dengue viruses, mainly in regions that are endemic for mosquito-borne viruses[1].

Arthropod-borne viruses are spreading worldwide, which may be due to migration and globalization[1]. Chikungunya outbreaks have been recorded in Africa and South America[1]. The virus was diagnosed in European countries, as well as in the USA[4] and Asia[5]. Out of all symptoms observed in Chikungunya infection, polyarthralgia, myalgia, and skin rashes are the most common[1]. Unexpected skin damage, such as nasal skin necrosis, have been observed[6]. Multiple aphtha-like ulcers have been described in the scrotal, penal, groin, perianal areas, and in the mouth[7]. However, no reports regarding genital lesions in women were found.

Multinucleated keratinocytes are cytological characteristics observed in herpes simplex, varicella-zoster, and measles infections. However, specific histopathological findings have not been developed for the Chikungunya virus. Riyaz et al. described nonspecific findings such as spongiosis, dermal edema, and perivascular lymphocytic infiltration in Chikungunya cases[8]. As these characteristics cannot be used for the final diagnosis, biopsies of the lesions were not performed on our patient.

The *Aedes Aegypti* mosquito transmits the virus when feeding on blood[9]. Human dermal fibroblasts, keratinocytes, melanocytes, and macrophages are susceptible to the virus[5]. In the presence of mosquito saliva, IFN-1 gene expression is decreased in fibroblasts[9]. The virus is cytopathic, which results in the apoptotic death of infected cells[5]. This mechanism explains the occurrence of skin ulceration on the external genitals, as well as other symptoms[5].

Table 1 – Work-flow of patient's laboratory tests for arboviral differential diagnosis.

Date	July 25, 2018	August 23, 2018	Laboratory normal ranges
Test	Result	Result	
Anti-Chikungunya IgM	<0.8	6.2	<1.1
Anti-Chikungunya IgG	<0.8	2.3	<1.1
Anti-Zika IgM	<0.8	<0.8	<1.1
Anti-Zika IgG	3.6	3.5	<1.1
Anti-Dengue IgM	0.13	0.15	<1.1
Anti-Dengue IgG	3.83	3.74	<1.1

It is possible for viral ulcers to heal in two weeks without recurrence[10]. Oral prednisolone or topical therapy can be used for symptom relief while anti-viral treatment is unavailable[10].

Strengths and limitations

This case report demonstrates a rare lesion on the vulva associated with viral disease. Due to the rarity of the condition, a large number of participants could not be included. However, case reports serve to develop hypotheses and accumulate knowledge in rare diseases.

CONCLUSION

This case serves to alert physicians to consider the possibility of mosquito-borne viruses among other potential causes when seeking a diagnosis for the etiology of vulvar ulcers.

Informed consent

The patient gave written informed permission to publish this case report and photos of the lesions after reading the final manuscript. Approval by the Human Research Ethics Committee.

Participation of each author

Isabel Cristina Chulvis Guimarães do Val consulted the patient, invited her to participate, made the final diagnosis, supervised manuscript writing, and reviewed the final manuscript.

Svitrigaile Grinceviciene drafted the manuscript, analyzed laboratory results, and participated in diagnostic evaluation.

Suzana Cristina Aidé Viviani Fialho evaluated the case and contributed to the final manuscript writing.

Renata do Val Guimarães analyzed laboratory results, participated in the diagnostic evaluation, and reviewed the final manuscript.

Guilherme Amaral Calvet analyzed laboratory results, conducted the differential diagnosis of viral infection, participated in diagnostic evaluation, and reviewed the final manuscript.

Funding

The present paper had no financial support.

Conflict of interests

The authors declare no conflict of interests.

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Received on: 02.28.2021
Approved on: 03.29.2021