Acta Scientific Gastrointestinal Disorders (ISSN: 2582-1091)

Volume 7 Issue 6 June 2024

Case Report

EBV-Induced Hepatitis and Juvenile Gangrenous Vasculitis: A Case Report

Karam Karam[1], Sarah Saleh[2], Houssein Chebbo[2], Rafca Challita[3], Antoine Salloum[4], Elias Saikaly[5], Walid obeid[1] and Elias Fiani[6]*

[1]*Department of Gastroenterology, University of Balamand, Koura, Lebanon*

[2]*Faculty of Medicine, University of Balamand, Koura, Lebanon*

[3]*Associate Professor of Rheumatology, Saint Georges Hospital UMC, Department of Internal Medicine, Lebanon*

[4]*Dermatology, Saint Georges Hospital UMC, Department of Dermatology, Dermatologic Surgicenter, Philadelphia, PA, United States*

[5]*Associate Professor of Surgery, Saint Georges Hospital UMC, Department of Surgery, Lebanon*

[6]*Gastroenterology, Associate Professor, Saint Georges Hospital UMC, Department of Internal Medicine, Lebanon*

***Corresponding Author:** Elias Fiani, Gastroenterology, Associate Professor, Saint Georges Hospital UMC, Department of Internal Medicine, Lebanon.

DOI: 10.31080/ASGIS.2024.07.0631

Received: May 06, 2024

Published: May 30, 2024

Abstract

Introduction: Juvenile gangrenous vasculitis (JGV) is a rare painful scrotal ulceration with less than 20 cases reported in the literature. The peak incidence is estimated between 5 and 45 years old. Patients usually report scrotal itching, stinging, burning or pain and these symptoms precede the development of lesions or ulcers. The etiology is unknown and in most cases no underlying infection can be identified

Case: A 29 years old male, previously healthy, presented for a three newly appearing scrotal ulcers. The patient was febrile 4 days ago and had sore throat, severe fatigue and decrease appetite without other gastrointestinal symptoms. His physical examination was pertinent for the presence of multiple pharyngeal ulcerations, a 1 cm retro-auricular adenopathy, with mild tenderness on the right upper quadrant and the presence of 3 active and painful scrotal ulcers. The EBV-VCA IgM was positive 160 U/mL (Negative < 20) confirming the presence of an acute EBV infection.

Discussion: A young patient presented with symptoms of pharyngitis, scrotal ulcer, increase in WBC, thrombocytopenia, and a perturbated liver enzymes. Workup for all viral hepatitis (A, B, C, EBV and CMV) was done to rule out hepatitis. The history of pharyngitis, the presence of thrombocytopenia, perturbated liver enzymes and hepato-splenomegaly on abdominal ultrasound was highly suggestive of EBV induced pharyngitis and hepatitis and was confirmed with the presence of a positive IgM EBV-VCA serology. A painful scrotal ulcer was noted suggestive of JGV.

JGV is a rare scrotal gangrene of unknown origin characterized by acute onset of skin ulcer and affecting mainly young patients. Notably, an association between LU and EBV is described in the literature, and this was reported too in our case. The disease is self-limited and a complete resolution within 2-3 weeks is noted.

Conclusion: EBV infection should be ruled out whenever a patient has disturbed liver enzymes, pharyngitis, hepatosplenomegaly and skin involvement, such as genital ulcers. Gastroenterologists should be prudent enough to exclude a diagnosis of JGV in context of EBV infection and genital involvement. JVG is a self-limited disease with a complete resolution in 2-3 weeks.

Keywords: Juvenile Grangrenous Vasculitis; Penile Ulcers; Genital Lesions

Introduction

Juvenile gangrenous vasculitis (JGV) is a rare painful scrotal ulceration with less than 20 cases reported in the literature [1,2]. Despite the fact that the disease dates back to 1974 (where it was described by Pinol., *et al*.) less than 20 cases are reported in the literature [1]. The peak incidence is estimated between 5 and 45 years old. Patients usually report scrotal itching, stinging, burning or pain and these symptoms precede the development of lesions or ulcers. The etiology is unknown and in most cases no underlying infection can be identified.

Owing to its rarity and the lack of a clear diagnostic criteria, the exact prevalence of the disease is still unclear. Only one case of JGV induced by EBV has been reported in the literature. Here we report a young male patient with EBV induced pharyngitis and hepatitis that developed multiple scrotal ulcers 4 days after the onset of his symptoms.

Case Presentation

A 29 years old male, previously healthy, presented for a new onset of three scrotal ulcers. The patient was febrile 4 days ago and had sore throat, severe fatigue and decrease appetite without other gastrointestinal symptoms. He denied local trauma, application of any product, sexual risk behavior, or similar previous episodes. His physical examination was pertinent for the presence of multiple pharyngeal ulcerations, a 1 cm retro-auricular adenopathy, with mild tenderness on the right upper quadrant and the presence of 3 active and painful scrotal ulcers. No inguinal lymph nodes were palpable. A Full blood workup was done and showed an increase in WBC of 13.200 x 10^6/L (N:4000-10000 x 10^6/L), with neutrophils of 47.3% and lymphocytes of 46.3%, a Hb of 14.4 g/dL (N: 13.5-17.5 g/dL) and low platelet count of 145000 x 10^6/L (N:150000-400000 x 10^6/L), a high CRP of 32 mg/L (N < 10 mg/L), liver enzymes were perturbated with an SGPT of 291 U/L (N <41), an SGOT of 171 U/L (N<37), a Gamma GT of 188 U/L (N: 11-49), a Phosphatase alkaline of 289 U/L (N: 40-129), with normal direct and total bilirubin, normal albumin, prothrombin time (PT) and partial thromboplastin time (PTT). The IgM antibodies against HSV 1/2, cytomegalovirus, parvovirus B19, and hepatitis A, were all negative. The workup for syphilis (Treponema pallidum particle agglutination assay test), human immunodeficiency virus (HIV), hepatitis B, and hepatitis C virus serology was also negative. HLA B 51 was negative. The EBV-VCA IgM was positive 160 U/mL (Negative < 20) confirming the presence of an acute EBV infection. Abdominal ultrasound showed a mildly enlarged liver span of 18 cm (N < 15) and a mild splenomegaly (18 x 13 x 12 cm).

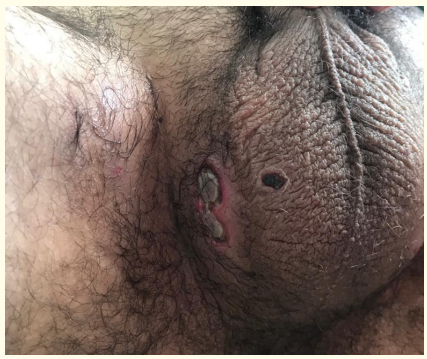

Figure 1: Multiple well demarcated scrotal ulcers.

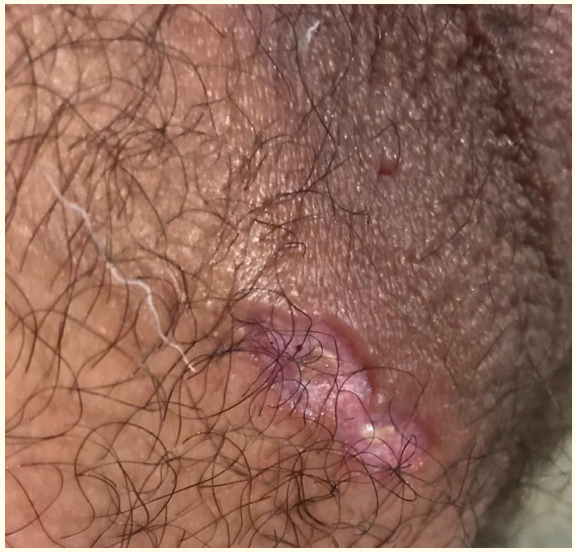

Figure 2: Well demarcated ulcer with mild pus noted in the center.

Discussion

A young patient presented with symptoms of pharyngitis, scrotal ulcer, increase in WBC, thrombocytopenia, and a perturbated liver enzymes. Workup for all viral hepatitis (A, B, C, EBV and CMV) was done to rule out hepatitis. The history of pharyngitis, the presence of thrombocytopenia, perturbated liver enzymes and hepatosplenomegaly on abdominal ultrasound was highly suggestive of

EBV induced pharyngitis and hepatitis and was confirmed with the presence of a positive IgM EBV-VCA serology. What was striking in this case is the presence of a painful scrotal ulcer suggestive of JGV.

JGV is a rare scrotal gangrene of unknown origin characterized by acute onset of skin ulcer and affecting mainly young patients [3]. It is hypothesized that JGV is the male counterpart of Lipschutz ulcers (LU). LU and JGV have similar clinical nuances; they occur in young patients, after prodromal symptoms (tonsillitis and fever), and consist of multiple tender necrotic ulcers [2]. Notably, an association between JGV and EBV is described in the literature, and this was reported too in our case [2]. In the literature, JGV was not linked to a specific drug, a systemic inflammatory, a rheumatologic or autoimmune disease, which was consistent with our case [2].

Pyoderma Gangrenosum (PG) has clinicopathological similarities with JGV [4]. However, PG is associated with systematic diseases, occurs in older population, spreads to extragenital areas, and requires medical treatment [5]. Therefore the consideration of a variant of PG was ruled out. JGV is self-limited and a complete resolution within 2-3 weeks is noted [2]. Notably, no relapse is reported, probably due to short follow-up duration [2,6].

Conclusion

This article elucidated a rare entity, Juvenile gangrenous vasculitis, induced by EBV infection. Gastroenterologists ought to be vigilant when consulted on a case of disturbed liver function tests in context of pharyngitis, hepato-splenomegaly and genital skin involvement to rule out an EBV infection. Furthermore, JGV should be differentiated from PG because they mimic each other given their clinico-pathological similarities. JVG is of unknown etiology and only 1 case report described the association between JVG and EBV infection. Nonetheless, JGV is a self-limited disease and resolves on its own with a complete resolution within 2-3 weeks.

Bibliography

1. Nettleton J., *et al*. "Scrotal necrosis and no Fournier's in sight: a rare case of juvenile gangrenous vasculitis". *BMJ Case Report* 12.2 (2019): e226530.

2. Chen W and Plewig G. "Lipschütz genital ulcer revisited: is juvenile gangrenous vasculitis of the scrotum the male counterpart?" *Journal of the European Academy of Dermatology and Venereology* 33.9 (2019): 1660-1666.

3. Caputo R., *et al*. "Juvenile gangrenous vasculitis of the scrotum: Is it a variant of pyoderma gangrenosum?" *Journal of the American Academy of Dermatology* 55 (2006): S50-53.

4. Marcos-Pinto A., *et al*. "Juvenile gangrenous vasculitis of the scrotum: A rare cause of genital ulcer". *Australas Journal of Dermatology* 61.4 (2020): e424-e426.

5. Satoh M and Yamamoto T. "Genital pyoderma gangrenosum: report of two cases and published work review of Japanese cases". *Journal of Dermatology* 40.10 (2013): 840-843.

6. Jimenez-Cauhe J., *et al*. "Juvenile gangrenous vasculitis of the scrotum: the male counterpart of Lipschütz ulcer?" *Journal of the European Academy of Dermatology and Venereology* 33.12 (2019): e459-e461.