ISSN: 2755-0117

Journal of Oncology Research Reviews & Reports

SCIENTIFIC Research and Community

Case Report

Open Access

Case Report: Hyperacute GVHD Presenting Solely with Genital Erosion in a 17-Year-Old Female

Amnah Almulhim*, Abeer Adel Alshamlan, Lenah Yassir Shaikh and Ayed Alqarni

Dermatology Department, King Fahad Specialist Hospital, Dammam, Saudi Arabia

ABSTRACT

We report a case of a 17-year-old Saudi female who developed hyperacute graft-versus-host disease (GVHD) presenting primarily with genital erosions. While genital GVHD has been documented in the literature, this case is unique as it presents with genital lesions as the primary and sole manifestation. We hope this report sheds light on other cases that might be underreported and emphasizes the importance of considering genital GVHD in similar patients.

***Corresponding author**

Amnah Almulhim, Dermatology Department, King Fahad Specialist Hospital, Dammam, Saudi Arabia.

Received: January 14, 2025; **Accepted:** January 18, 2025; **Published:** January 25, 2025

Introduction

Vulvovaginal GVHD is a subtype of GVHD that affects the vulvovaginal region. It is common; however, it is often underreported and underdiagnosed in female patients [1]. We report a unique case of a young female with hyperacute GVHD as the primary manifestation.

Case Presentation

We describe a 17-year-old single female with GDPD deficiency and an insignificant family history of malignancies (she has two brothers and one sister). She was diagnosed with stage IIIB small cell carcinoma of the ovary, hypercalcemic type, in 2021, for which she underwent fertility sparing surgery (debulking laparotomy, left salpingo oophorectomy, omentectomy, appendectomy, and aortic lymph node dissection) in January 2022. A total abdominal hysterectomy with right salpingo oophorectomy and pelvic and para-aortic lymph node dissection was performed in March 2022. She was later diagnosed with acute myeloid leukemia (AML) in May 2024 and received chemotherapy with cisplatin, etoposide, and bleomycin, followed by treatment for therapy-related AML. An allogeneic stem cell transplantation (SCT) was performed on September 23, 2024, with her brother as the donor. Her post-transplant course was complicated by multiple issues, including febrile neutropenia and grade 3 oral mucositis. Thirteen days after the transplant, she developed a progressive skin rash in the genital area, accompanied by a burning sensation but no itching. Pain improved with the application of topical lidocaine. She also experienced fever, nausea, vomiting, and diarrhea starting with the onset of the genital rash. A few days later, seventeen days post-transplant, she developed bilateral areolar and nipple erosions, which were painless and non-itchy. Additionally, she reported unilateral sole pain and skin nodules, with no previous history of similar rashes. The primary team initially diagnosed her with vulvar cellulitis and disseminated herpes simplex infection, she received systemic antibiotics and antiviral for 7 days; however, the rash did not resolve.

A few days later, she began to exhibit elevated liver enzymes, raising suspicion of GVHD. At that time, we were consulted, under examination the labia majora showed hypopigmented patches with mild atrophy over the mucosal surface of the labia and perianal area with mild watery discharge, and multiple fresh erosions without discharge. There were also well-defined areolar and nipple erosions with dry scales and exfoliation.

Due to the unclear clinical picture and suspicion of GVHD, a decision was made to perform a biopsy. A punch biopsy of the labia majora revealed mild epidermal spongiosis with scattered pyknotic apoptotic keratinocytes, predominantly at the basal portion of the epidermis. Minimal perivascular lymphocytic infiltrate was noted in the superficial dermis, along with mild ectatic dermal vessels and pigment incontinence. The deeper portion of the dermis and skin adnexa were essentially unremarkable, with no viral changes or specific pathogens identified (Figure 1).

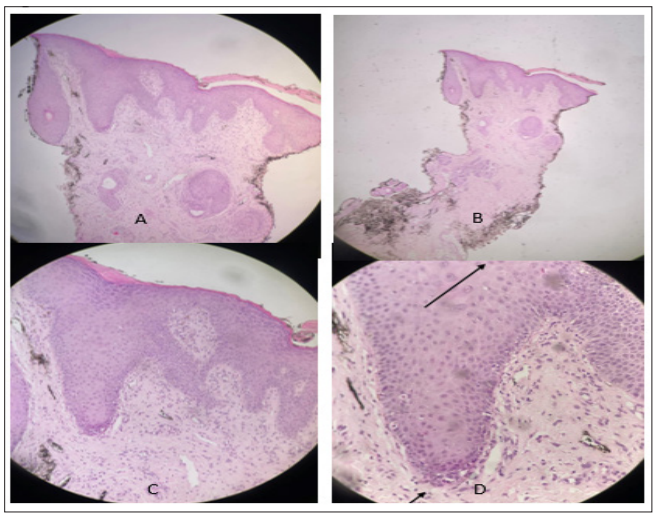

Figure:1

Following the biopsy results, a conclusion was drawn based on the histopathological findings, which were consistent with acute graft versus-host disease.

Discussion

Genital GVH is well recognized and described in the literature, several cases has been reported, recognizing it is based mainly on clinical symptoms and examination but due to the sensitivity of the area it is commonly not examined and patients might be hesitant to report symptoms specially in female young individuals. Failure to diagnose and treat such cases can lead to sclerotic changes in the genital area and complete vaginal obstruction [1,2]. Moreover, a delay in the diagnosis of GVHD can result in a more exaggerated and severe presentation. A systematic review conducted in 2023, which included 32 young females under 20 years of age, concluded that 83% developed grade 3 genital GVHD, with the time of diagnosis ranging from 62 to 2966 days, and a median of 381 days post-transplant [3]. Risk factors for GVHD include HLA incompatibility, female donor to male recipient, unrelated donor, and the source of stem cells, with the highest risk associated with peripheral blood use [4]. Reported symptoms include dryness, vulvar pain, itching, dysuria, dyspareunia, vulvar adhesions, loss of architecture of the labia minora and clitoris, and erosions. Conversely, some patients may be completely asymptomatic at the time of diagnosis [2,3]. This report presents a unique case of a young female with a complex oncological background who developed hyperacute GVHD characterized by genital and peri areolar erosions, with no identified risk factors. We emphasize the importance of considering genital GVHD in all HSC transplant patients, as patients may not report symptoms due to the sensitivity of the area, and not being commonly examined in similar cases.

Conclusion

We recommend inquiring about genital involvement and examining the area whenever feasible. Delay in diagnosis may lead to more severe manifestations of GVHD and complicate treatment, as additional organs may become involved. We hope this case aids in the early diagnosis of unrecognized instances of GVHD.

Reference

1. Jacobson M, Wong J, Li A, Wolfman WL (2019) Vulvovaginal graft-versus-host disease: a review. Climacteric 22: 603-609.
2. Cizek SM, El-Bietar J, Rubinstein J, Dandoy C, Wallace GH, et al. (2019) Pediatric and Young Adult Vulvovaginal Graft-versus-Host Disease. Biol Blood Marrow Transplant 25: 2408-2415.
3. Tsimeki M, Tsimpidakis A, Roidi S, Gregoriadis T, Soldatou A, et al. (2023) Genital GVHD in Female Children and Adolescents: A Systematic Review of Case Reports and Case Series. Children (Basel) 10.
4. Gratwohl A, Sureda A, Cornelissen J, Apperley J, Dreger P, et al. (2017) Alloreactivity: the Janus-face of hematopoietic stem cell transplantation. Leukemia 31: 1752-1759.