

Unusual Presentation of Kaposi Sarcoma During Adalimumab Therapy: a Case Report

Gulsun-Hazan Tabak¹, Basak Yalici-Armagan¹, Burkay-Adem Sahin¹, Ozay Gokoz², Sedat Kiraz³

1 Hacettepe University, School of Medicine, Department of Dermatology and Venereology, Ankara, Turkey

2 Hacettepe University, School of Medicine, Department of Pathology, Ankara, Turkey

3 Hacettepe University, School of Medicine, Department of Rheumatology, Ankara, Turkey

Key words: psoriasis, biologics, kaposi sarcoma, TNF antagonist

Citation: Tabak GH, Yalici-Armagan B, Sahin BA, Gokoz O, Kiraz S. Unusual Presentation of Kaposi's Sarcoma During Adalimumab Therapy: A Case Report. *Dermatol Pract Concept.* 2022;12(3):e2022071. DOI: https://doi.org/10.5826/dpc.1203a71

Accepted: October 11, 2021; Published: July 2022

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Funding: None.

Competing interests: None.

Authorship: All authors have contributed significantly to this publication.

Corresponding author: Basak Yalici-Armagan, MD, Hacettepe University, School of Medicine, Department of Dermatology and Venereology, 06100, Ankara, Turkey, Telephone: 00903123051704, Fax: 0 3123114340, E-mail: basakarmagan@gmail.com

Introduction

Kaposi sarcoma (KS) is a rare angioproliferative malignancy associated with Human Herpesvirus-8 (HHV-8) that involves skin and visceral organs. It typically manifests with magenta-colored macules, papules, and nodules on the skin. Herein, we present a patient who developed pyogenic granuloma (PG)-like KS during adalimumab therapy for psoriatic arthritis.

Case presentation

A 48-year-old male applied to our dermatology outpatient clinic for a bleeding lesion on the right forth toe. The lesion occurred seven-months before and grew rapidly in last onemonth. He has been diagnosed with psoriatic arthritis 4 years ago and treated with adalimumab, a tumor necrosis factor- α inhibitor (TNFi), for the last one year. Dermatological examination revealed a 1cm x1 cm diameter of reddish, ulcerated, protruding hemorrhagic nodule partially covered with hyperkeratotic crust and fibrin just under the fourth toe nail (Figure 1, A and B). Excisional biopsy was performed with provisional diagnoses of amelanotic malignant melanoma, PG and cutaneous squamous cell carcinoma. Histopathological examination showed nodular lesion in the dermis made up of atypical spindle shaped cells showing whorled structures and slit like spaces filled with erythrocytes. Immunohistochemical examination indicated HHV-8 positivity and confirmed the diagnosis of KS (Figure 1E). In detailed dermatological examination, purple-colored patches on the lateral aspects of both feet were noticed (Figure 1, C and D). HIV serology was negative. Adalimumab treatment was ceased immediately and the patient was addressed to oncology department for remaining lesions.

Conclusions

The potential of TNFis to increase the risk of malignancy including melanoma and non-melanoma skin cancer is still controversial. Although iatrogenic KS has been described in



organ transplant recipients receiving immunosuppressive therapy, there are only a few reports of KS during TNFi therapy that were presenting as typically purplish-red patches and papulonodular lesions on the lower leg [1]. Classic KS is typically seen in older men (64-72 years) of Mediterranean, Eastern European (Ashkenazi) Jewish, or South American origin whereas endemic KS is limited to sub-Saharan Africa and is typically seen in young (25-40 years), black, HIV-negative men. On the other hand, KS and psoriasis speculated to share common pathogenesis particularly related with interleukin-6 cytokine pathway or the same human leukocyte antigen alleles. Although we cannot explain the exact mechanism of this association whether it is a co-existence of KS and psoriasis or it is triggered by adalimumab, HIV negativity and the early age of onset compared to classical KS suggested a relationship with adalimumab in the present case. Unlike reported KS cases induced by TNFi treatment, PG-like presentation with classical purple-colored patches was a remarkable finding of the current case. PG-like KS is a rare variant of KS that is difficult to diagnose clinically [2]. We present this rare case of PG-like KS that can be easily confused with other skin tumors in a patient under biologic treatment.

If characteristic purple-colored patches of KS on the lateral sides of feet were not overlooked on physical examination in the current case, it could have been possible to include KS in the clinical pre-diagnoses. Detailed whole body skin examination is crucial for dermatologists to provide correct clinical diagnosis for early management. Further evidence is needed to clarify the relationship between biologics and malignancy development.

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