Case Report
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Bullous kaposi sarcoma induced by rituximab for leukemia. case report

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Introduction

Kaposi sarcoma (KS) is a multifocal low-differentiated mesenchymal angioproliferative tumor caused by KS associated human herpesvirus type 8 (HHV8). Four variants of KS - Epidemic or AIDS-associated KS, Endemic or African KS, Iatrogenic KS and Mediterranean or classic KS (CKS) are described.

Blue -purple plaques and nodules are common for KS patients, on rare occasion, growths, granulations, vesicles and bullas are sometimes observed. Several clinicopathologic variants of KS have been described: regular, anaplastic, lymphangiectatic, hyperkeratotic, micronodular, telangiectatic, keloidal, etc.

Histologically KS lesions are comprised of spindle shaped tumor cells, abnormal vessels and a variable chronic lymphoplasmatic inflammatory infiltrate. KS lesions grow from an early patch stage, to form plaques, which ultimately develop into a nodular (tumor) stage. Bullous changes in KS are rarely encountered in real practice[1] and the main pathological sign of bullous KS is intraepidermal or subepidermal splitting.[2]

KS occurs most commonly in immunocompromised individuals such as HIV-infected patients. Iatrogenic Kaposi sarcoma is developed in patients as a result of immunosuppressive therapies for autoimmune disorders or after organ transplantation.[3] Corticosteroids, methotrexate, cyclophosphamide, azathioprine, biologicals and many other drugs that can suppress immune system have been associated with KS.[4,5] Inflammation and ~~an~~ insufficient T-cell surveillance, primarily due to genetic predisposition, iatrogenic immune suppression or co-infections appear to be the most important cofactors. According to our literature search four publications reporting ~~of~~ KS association with rituximab treatment were found.[6–9]

Rituximab is a chimeric murine/human monoclonal IgG antibody (mAb) targeted against the CD20 antigen present on the surface of mature B cells. Rituximab is now widely used in the treatment of many dermatologic and rheumatologic conditions. Since its approval for relapsed/refractory non-Hodgkin's lymphoma in 1999, rituximab has been licensed for the treatment of other numerous B-cell malignancies, including the follicular lymphoma, as well as autoimmune conditions, including rheumatoid arthritis.[10] The use of Rituximab is associated with some side effects and increased risk of infections such as tuberculosis[11] or autoinflammatory conditions such as pyoderma gangrenosum[12] and psoriasis.[13]

Herein we report the case of Kaposi sarcoma diagnosed in a HIV-negative patient under rituximab treatment for her leukemia, as the first case of rituximab-induced bullous KS.

Case report

An 83-year-old Caucasian woman known to have B-cell chronic lymphocytic leukemia (CLL) since 2002 presented with painful rash on her left leg accompanied by general weakness. In 2018 due to the severity of her CLL manifestations (progressive weakness, splenomegaly, frequent viral infections) treatment with rituximab 375 mg/m2 was started once a week for 5 consecutive weeks as definitive treatment for CLL. After 5 weeks of rituximab initiation a violet-purple rash appeared on her left leg. The patient continued her treatment with rituximab for CLL. Later on, because of progressive splenomegaly and lymphadenopathy Chlorambucil was added. Meanwhile the skin rash continued to progress. On admission in dermatology department blue-purple macular, papular and bullous lesions accompanied by hyperkeratotic and papillomatoses painful overgrowths primarily on the anterior aspect of her left leg were seen (Figure 1). After a dermatological evaluation, the lesions were biopsied. The pathologic work up revealed a bulla in hyperkeratotic epidermis.

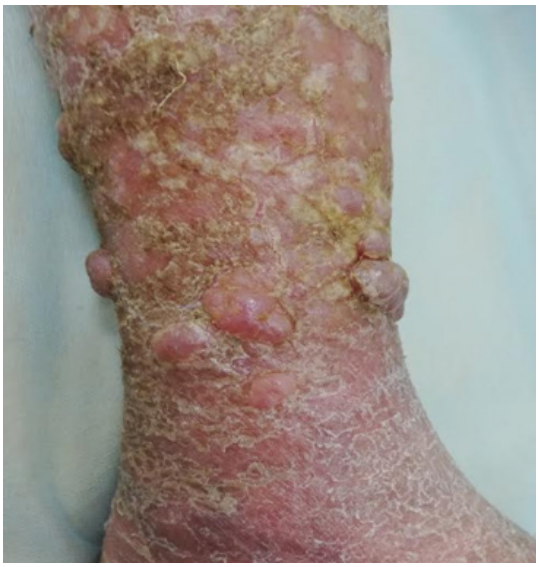

Figure 1 Blue-purple nodules, papillomatoses overgrowths and tight bullous lesions.

Acanthosis is not expressed; dermal papillae are focally smoothed. Irregular bizarre vascular spaces dissecting between collagen fibers in the mid and deep dermis are observed. Entire dermis is filled with a diffuse proliferation of fascicular spindle cells. The specific vascular marker in skin CD34 is positive. The skin and blood are immunoreactive for HHV-8. The pathology concluded a diagnosis of Bullous Kaposi sarcoma (Figure 2).

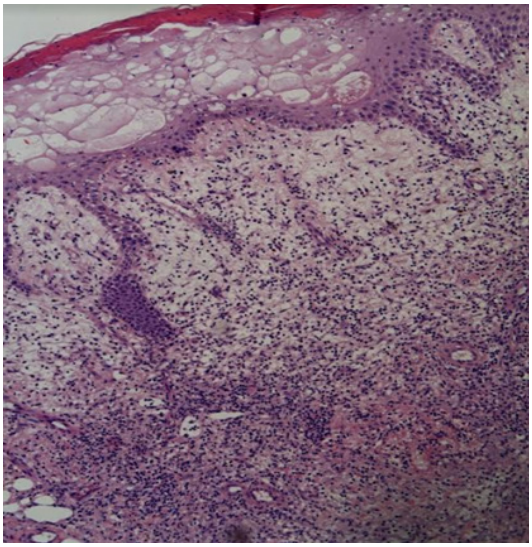

Figure 2 Skin biopsy: an intraepidermal bulla overlying mild subepidermal lymphedema with irregular bizarre vascular spaces in the mid and deep dermis. (H&E stain) X100.

Conclusion

This is the fifth case report describing rituximab-induced Kaposi sarcoma and the first one of its bullous form. The described case further highlights the crucial role of immunosuppression in the pathogenesis of Kaposi sarcoma.

Acknowledgments

None.

Conflicts of interest

Authors declare there is no conflict of interest.

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