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Case Letter

# Iatrogenic Kaposi's sarcoma in the anovulvar area

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Sir,

Kaposi's sarcoma (KS) is an indolent angioproliferative tumor that depends on viral replication (HHV-8) and inflammatory cytokines produced by infected immune and endothelial cells [1]. It is a multifocal disease, with its evolutionary spectrum varying from a locoregional "indolent" form to a disseminated and fulminant form. KS lesions are rarely limited to unusual mucocutaneous areas. To our knowledge, there have been no reports of iatrogenic KS confined to the female external genitalia. Herein, we report the first case of iatrogenic KS restricted to the vulva and anus in an HIV-negative patient.

An 85-year-old female presented with violaceous nodules on the external genitalia. Her past medical history was significant for post-hepatitis C cirrhosis treated with sofosbuvir and daclatasvir for six months (the viral load of the control was negative). The patient developed bullous pemphigoid two weeks after the initiation of the anti-viral treatment. Thereafter, corticotherapy was started at the dose of 1 mg/kg/day. During follow-up, the patient had a complete healing of the pemphigoid lesions. On control, four months after the initiation of corticotherapy, a physical examination revealed several brownish-violaceous angiomatous nodules and slightly raised plaques on both labia majora and minora reaching up to the anus. The labia majora were swollen and painful with lymphatic edema (Figs. 1 and 2). No other significant mucocutaneous lesions were observed, and there was no evidence of inguinal lymphadenopathy. Fibroscopy showed no involvement of the digestive mucosa. A chest X-ray and abdominal and pelvic CT showed no visceral lesions. HIV serology was negative. A histological study revealed dermal proliferation of spindle cells, slit-like vascular spaces, and extravasated red blood cells. A PCR assay revealed HHV-8 DNA sequences in the lesional skin tissue. For our patient, we suggested bleomycin intramuscularly 5 mg per day for three days in a row every four weeks, and a faster reduction in corticosteroid therapy.

Kaposi's sarcoma (KS) is a spindle-shaped vascular cell tumor that may be located in the skin, the gastrointestinal and respiratory tract, or the lymphoid organs [2]. Four types of histologically indistinguishable KS exist: classic, endemic, immunosuppressive-therapy-related, and epidemic [1]. Each form seems to be connected with HHV-8 infection.

The iatrogenic form is caused by immunosuppressive drugs used after an organ transplantation. This form may also appear following the use of systemic corticosteroids and dermocorticoids [3]. The majority of cases of KS limited to the external genitalia are observed in males, especially on the penis. In females, it is highly uncommon and is observed mainly in HIV-positive females (up to five times more frequently than in HIV-negative females) [4,5]. In fact, to our knowledge, one case of classic KS confined to the female external genitalia not associated with HIV infection has been reported [6]. Clinically, it presents itself as a tumor mass, papilloma, or abscess. Confirmation of the diagnosis is based on biopsy and virological tests showing the presence of KSHV in the lesional tissues [4,5]. Cryotherapy, surgical excision, and radiotherapy are the main local treatments in the case of solitary lesions. The use of interferon alpha has also produced good results. In the case of iatrogenic MK, there is no uniform treatment regimen for KS. Immunosuppression must be reduced to the lowest levels while preserving allograft function in the case of an organ transplantation. Cyclosporine A should

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**Figure 1:** Edematous labia with several brownish-violaceous nodules and slightly raised plaques on both the labia majora and minora.

**Figure 2:** Brownish-violaceous nodules and slightly raised plaques around the anus.

be converted to mycophenolate mofetil or mTOR inhibitors. Sirolimus appears to inhibit the growth of established vascularized tumors and this effect is best achieved with relatively low immunosuppressive doses of the drug [2].

Kaposi's sarcoma must be considered in the case of vulvar or anal tumor masses. Strict skin surveillance is necessary in patients treated with corticosteroid therapy or other immunosuppressive drugs.

## Consent

The examination of the patient was conducted according to the principles of the Declaration of Helsinki.

The authors certify that they have obtained all appropriate patient consent forms, in which the patients gave their consent for images and other clinical information to be included in the journal. The patients understand that their names and initials will not be published and due effort will be made to conceal their identity, but that anonymity cannot be guaranteed.

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