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Case Report
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Kaposi's Sarcoma Complicating Crohn's Disease in an HIV-Negative Patient: Case Report

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Abstract

Kaposi's sarcoma is a vascular proliferative disease associated with human infection with herpesvirus-8. It is more common in people with an immune deficiency, mainly those with Human Immunodeficiency Virus infection (HIV). Taking immunosuppressants or glucocorticoids in the context of inflammatory bowel diseases may in rare cases cause Kaposi's sarcoma in patients without human immunodeficiency virus. This association, although rare, causes a real diagnostic and therapeutic challenge.

This article reports a case of Kaposi's cutaneous sarcoma in a 64-year-old patient not infected with HIV, followed for Crohn's colonic disease treated with glucocorticoids and Azathioprine at the gastroenterology department of the CHU Mohamed VI in Marrakech.

The aim of this article is to encourage clinicians to adopt a multidisciplinary approach to diagnosis and treatment, so that rare cases can be diagnosed and treated more accurately than and as quickly as possible.

Keywords: Kaposi's sarcoma; Inflammatory bowel disease; Human herpesvirus-8; Immunomodulatory therapy; Crohn's disease

Introduction

Kaposi's sarcoma (Kaposi's sarcoma, SK) is a vascular proliferative disease associated with infection with Human Herpesvirus-8 (HHV-8), and is common in immunocompromised people. Kaposi's sarcoma is relatively rare, and occurs mainly in people infected with the Human Immunodeficiency Virus (HIV). In fact, in addition to the well-known HIV related Kaposi sarcoma, there is another type of Kaposi sarcoma, namely iatrogenic Kaposi sarcoma, which is mainly related to It is related to the use of immunosuppressive agents, which is the case with our study. The essence of inflammatory bowel disease is inflammatory bowel disease of unknown etiology, which is closely related to immune function. Skin manifestations may appear as extra digestive manifestations of the disease and others as drug-related complications. Because of these factors, the treatment of skin lesions varies depending on the pathophysiological mechanism involved. The problem is then to distinguish between these two types of lesions to ensure optimal patient care.

This article reports a case of cutaneous Kaposi's sarcoma in a patient with Crohn's disease treated with glucocorticoids and immunosuppressive therapy and reviews the literature.

Case Presentation

This is the case of a 64-year-old man followed since 2008 for colonic Crohn's disease of inflammatory phenotype without anoperineal lesions revealed by a mucous-bloody diarrhea at a rate of 3 to 4 stools per day with rectal syndrome without Koenig's syndrome initially treated with oral corticosteroids associated to 5-aminosalicylates. Symptoms were relieved after treatment with Mesalazine. The patient remained in clinical remission on Mesalazine 2 g/day for 10 years. In 2017 the patient presented a moderate outbreak controlled by oral corticosteroids then the patient was put on azathioprine 2.5 mg/kg/day with good evolution. In September 2018 the patient presented with purplish nodular lesions pruritic of all the body while the patient was in luminal remission 1 stool per day. The patient was hospitalized in the dermatology department with clinical examination of the patient who objectified confluent erythematous-purplish papules covering approximately 90% of the surface of the body, sparing the palms of the hands and the soles of the feet with white ichthyosiform scales (Figure 1, 2). In the biological assessment white blood cells 8,490, hemoglobin

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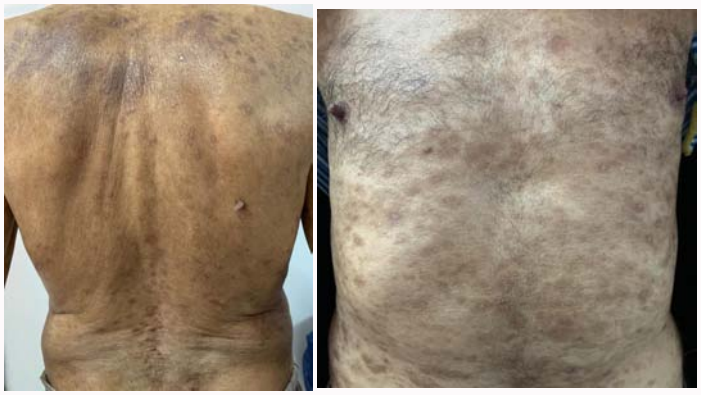

Figure 1 and 2: Purplish papules on the back and chest covered with ichiotiform scales in places.

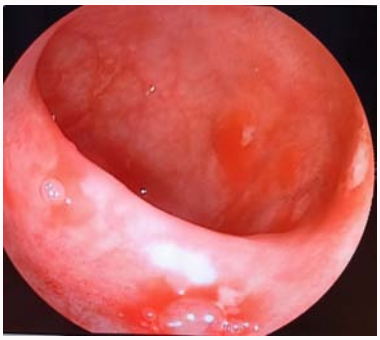

Figure 3: Colonoscopic appearance of erythematous mucosa with some superficial ulcerations.

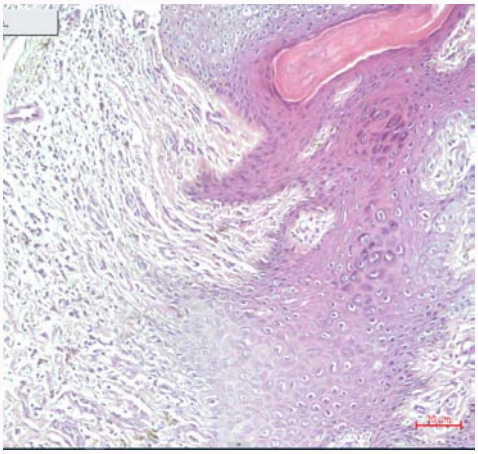

Figure 4: Lymphocytic dermatitis with marked melanin pigmentary incontinence and cell turgor.

13.3 g/dl, platelets 237,000. With the blood smear less than 4% of Sezary cells were present. Red blood cell sedimentation rate 3 mm/h, C-reactive protein 3.1 g/L, glycemia at 0.88 g/l, hepatitis B and C serologies, HIV, syphilis, were negative. A colonoscopy was performed showing a pseudopolypoid erythematous mucosa without ulcerations (Figure 3) with histological study of subacute and chronic inflammatory changes without signs of acute infection without CMV inclusions. The other stool pathogenicity tests were negative, immunoglobulin M was negative. A skin biopsy of the lesions was carried out showing lymphocytic dermatitis with marked melanin pigmentary incontinence (Figure 4) with demonstration of anti

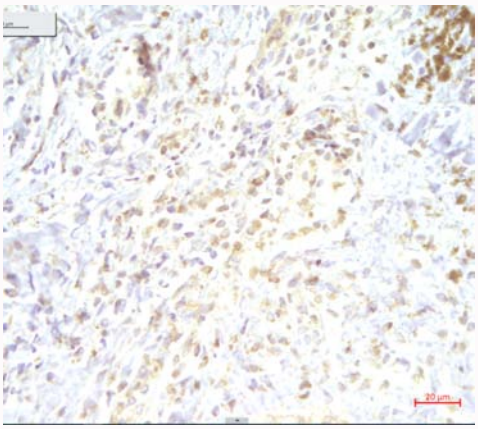

Figure 5: Staining of the nuclei of cells containing HHV8 antigen with stained anti HHV8 antibodies.

HHV8 appearance evoking Kaposi's sarcoma (Figure 5).

Discussion

HHV-8 infection is the underlying cause of all four epidemiologic forms of KS [1]. The prevalence of HHV-8 infection varies widely by geography, race, ethnicity, and risk group. The highest prevalence of HHV-8 is found in groups with a high incidence of KS, such as sub-Saharan Africans, individuals from Mediterranean countries, and individuals with HIV infection [2]. HHV-8 is mainly transmitted sexually, with the possibility of parenteral transmission (blood transmission, organ transplantation) and horizontal transmission mainly in highly endemic areas. It should be noted that our patient did not belong to any of the risk groups described above. In Spain, the seroprevalence of HHV-8 ranges from 0% in children to 6.5% in blood donors [3]. There are limited data on HHV-8 infection in patients with IBD [4]. Seventeen on the other hand, in a group of 60 patients with Crohn's disease receiving immunosuppressant treatment with infliximab, the presence of HHV-8 could not be demonstrated by serum PCR either [5]. The combination of HHV-8 and HIV increases the risk of developing KS by 30% within 10 years [6]. Although the risk of KS in IBD patients receiving immunomodulatory therapy remains very low, knowledge of HHV-8 serologic status is important in defining those at risk for developing KS. Iatrogenic KS is associated with immunosuppressive therapy. Similar to other viruses of the herpes family, HHV-8 remains latent after primary infection and can be reactivated in immunocompromised people. It has recently

been observed that a lytic viral gene encoding vGPCR (G Protein Coupled Receptor) plays a key role in the genesis of KS [7,8]. A finding that supports the important role of immunosuppression in the development of KS is that characteristic skin lesions regress after discontinuation of immunosuppressive therapy in both renal transplant patients [9] and in patients with UC, despite the persistence of HHV-8 DNA [10]. Currently, the diagnosis of KS requires clinical and histological criteria; however, knowledge of its association with HHV-8 has made detection of this infection essential for the management of these patients. Immunohistochemistry was used to localize HHV-8 proteins and to assess the involvement of HHV-8 in malignant tumors. Thus, the specific antigenic expression of HHV-8, particularly the detection of the latent nuclear antigen [11], allowed the diagnosis of KS in tissue samples [12]. In our case, HHV-8 immunohistochemistry was critical in confirming SK. With this case, we would like to highlight the importance of the use of immunomodulatory therapy in patients with chronic inflammatory bowel disease in relation to the development of KS. This tumor is associated with infection with HHV-8; therefore, this virus should be included among the possible opportunistic infections associated with immune suppression in patients with IBD. Since skin involvement is infrequent and clinical and endoscopic findings may be similar to inflammatory disease itself, a high degree of clinical suspicion is required to diagnose these patients.

Conclusion

Cutaneous Kaposi's sarcoma complicating chronic inflammatory bowel disease on immunosuppressant's remains, although rare, a serious condition that must be managed with a conservative approach and discontinuation of immunosuppressive therapy. It is therefore wise to keep in mind that this treatment-related complication can occur even during the first months of treatment, in order to be able to act quickly and correctly.

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