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(CASE STUDY)

HIV-Negative Gastric Kaposi's sarcoma: A Case Report

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Abstract

Kaposi's sarcoma is a mucocutaneous disease that mainly affects the elderly and HIV-infected individuals. However, digestive localization has often been observed during upper digestive endoscopies performed to search for secondary localization. It most often occurred in non-immunocompetent patients. However, endoscopy remains the examination of choice for confirming digestive tract involvement in KS. KS is diagnosed histologically, but in the literature, only 15-20% of endoscopic biopsies are positive for KS. We report a gastric case of Kaposi's sarcoma patient with negative HIV serology who was referred to a digestive endoscopy unit in search of secondary digestive lesions.

Keywords: Kaposi's sarcoma; Kaposi's disease; HIV negative; Upper gastrointestinal fibroscopy; Histopathology

1. Introduction

Kaposi's sarcoma is a mucocutaneous disease that mainly affects the elderly and HIV-infected individuals. The risk of developing Kaposi's sarcoma during AIDS is estimated to be 20,000 times that of the general population and almost 300 times that of other immunocompromised subjects [1]. The examination of choice for the diagnosis of digestive involvement is upper gastrointestinal fibroscopy. Histopathology is the gold standard for diagnosis. Few works have been devoted to digestive KS in Africa, and in particular to non-HIV-related KS, hence the interest of this observation. We report a gastric case of Kaposi's sarcoma patient with negative HIV serology who was referred to a digestive endoscopy unit in search of secondary digestive lesions.

2. Observation

A 78-year-old woman was admitted to the dermatology department with the appearance of a lesion on the left foot. On clinical examination, the lesion was an erythematoviolaceous papulo-nodule on the left foot, covered with hyperkeratosis. It was hard and painful to the touch, and associated with lymphedema (Figure 1). Histopathological examination after skin biopsy revealed a dermis infiltrated by a tumor proliferation of fasciculate architecture, with variable-sized, sometimes narrow, anastomosing fissures containing red blood cells. Tumor cells are spindle-shaped, sometimes with cytoplasmic vacuolization. These cells show moderate atypia. Immunohistochemistry revealed positive anti-HHV8 antibodies in tumor cells, suggesting Kaposi's sarcoma (Figure 2). HIV serology was negative on two occasions. As part of the search for digestive involvement, an upper gastrointestinal fibroscopy was performed, revealing an erythematous fundic mucosa with millimetric angiomatous flat lesions and normal bulbar mucosa, with a polypoid angiomatous lesion measuring approximately 5 mm on the roof (Figure 3), which was biopsied with results favoring a gastric localization of Kaposi's disease. The patient underwent 6 courses of Bleomycin with extension of the plantar lesions, and was then referred to an oncology department.

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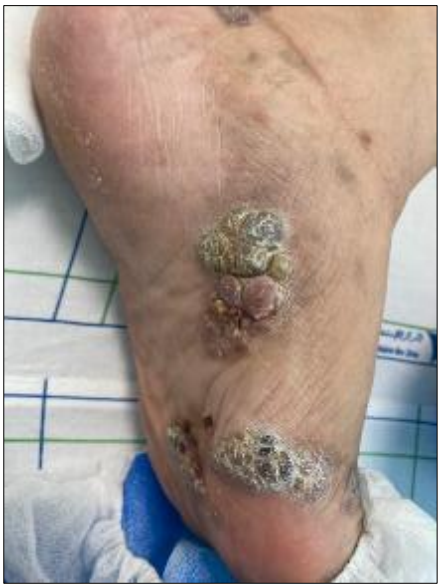

Figure 1 Plantar skin lesions

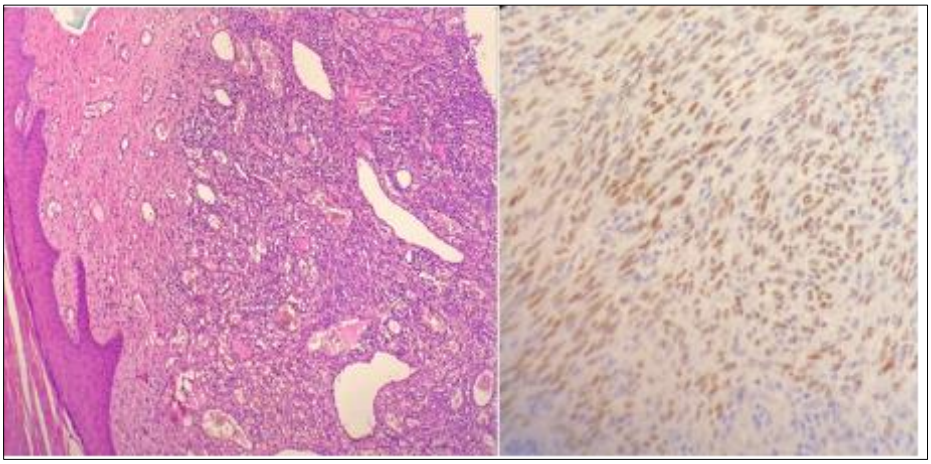

Figure 2 Histopathological appearance of Kaposi's sarcoma + Immunohistochemistry

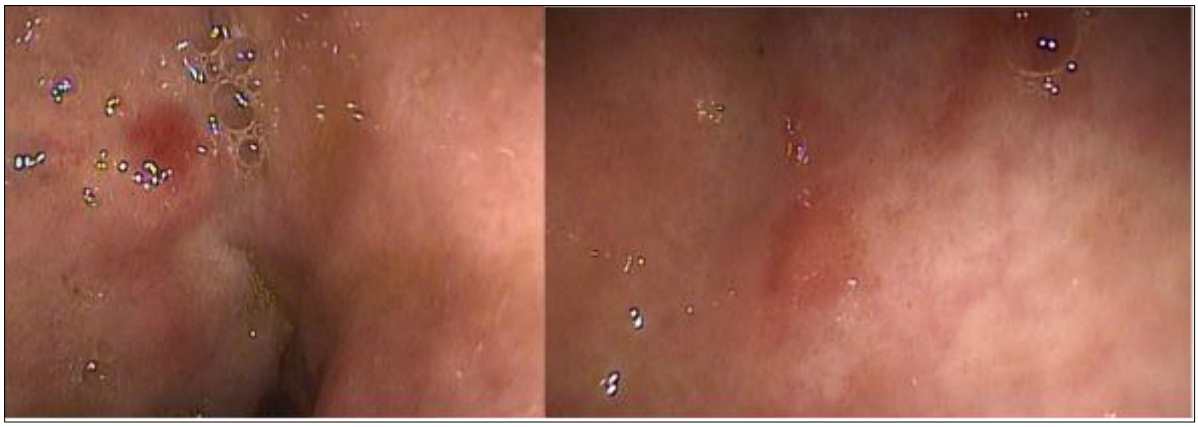

Figure 3 Endoscopic appearance on the left: the bulbar lesion, and on the right: the fundic lesion

3. Discussion

Kaposi's sarcoma (KS) can be defined as a mesenchymal proliferative process involving cells of the blood and lymphatic systems, which is induced by viral growth factors, notably interleukin 6 from human herpes virus type 8 (HHV8) [2]. Identified in 1872 by the Austrian dermatologist Moritz Kohn Kaposi, KS has long presented in two forms, one sporadic, cutaneous, or classical KS, the other endemic in Central Africa, or African Kaposi's, described in 1950 in Central and East Africa, a disease of adults and children [1]. Until the advent of Acquired Immune Deficiency Syndrome (AIDS), the disease was rare, with a slow and uncertain course. Four forms of KS have been identified: a classical Mediterranean form, described in 1872 by Moritz Kaposi in Vienna, Austria: a rare, malignant cutaneous disease affecting mainly elderly men; an endemic form described in 1950 in Central and East Africa, around the Great Lakes: a so-called post-transplant form linked to iatrogenic acquired immunodepression, described in 1970 in transplant patients treated with immunosuppressants and an AIDS-related epidemic form, described in 1981, in HIV-infected individuals[2]. Digestive involvement is rare in the classic, endemic form, but more frequent in the AIDS-related form. In this report, we present data on the gastroscopic findings in a newly recognized endemic form of KS in a defined Mediterranean population [3-4]. This form of KS is not related to HIV infection and usually occurs in the elderly as a systemic malignant disease. This is a very specific anatomical and clinical entity, with cutaneous localizations dominating the picture, and usually confined to the lower limbs. Involvement of the digestive tract is rare [5]. The elementary lesion is usually a macule which evolves into a papule (oval, 0.5 to 1cm in diameter, poorly limited, purplish-brown, classically painless, not pruritic); a nodule (hard, mobile); a placard (angiomatous, edematous, sometimes painful); an ulcero-vegetative tumor, sometimes sessile or pediculated. Whatever the initial lesion, it is well limited, angiomatous, erythematous then purplish, appearing hyperpigmented in relation to the underlying skin [6]. Digestive localizations are usually asymptomatic, and rarely cause digestive hemorrhage or perforation. [6]. However, digestive localization has often been observed during upper digestive endoscopies performed to search for secondary localization. It most often occurred in non-immunocompetent patients. The endoscopic appearance of gastric KS was first described by Rajah et al. in 1969,27 but to date the published information on the prevalence of gastrointestinal manifestations in KS is variable [7]. Digestive involvement almost always occurs in association with skin lesions. The esophagus, stomach, duodenum, colon and rectum may all be affected. In most cases, involvement of the digestive tract is asymptomatic. However, endoscopy remains the examination of choice for confirming digestive tract involvement in KS. Initial lesions may be flat. At a more advanced stage, symptomatic lesions usually appear as red, sessile, protruding nodules, sometimes ulcerated. These are polypoid, lenticular or oval formations, from a few millimeters to 1 or 2 cm in diameter or more, isolated or contiguous, enclosed in fine fibrin networks giving them a squared appearance. Their color ranges from bright red to wine-red. Biopsies are often negative, as the tumor is located in the submucosa. In the literature, only 15-20% of endoscopic biopsies are positive for KS. KS is diagnosed histologically, as a proliferation of irregularly anastomosing vascular clefts, insinuating themselves between collagen clusters and extending around normal dermal vessels and adnexa, mixed with spindle cell bundles, a predominantly lymphocytic mononuclear inflammatory infiltrate with extravasated red blood cells taking on the appearance of hyaline globules once phagocytosed. HHV8 immunostaining is positive [2]. Extracutaneous involvement is very common in HIV-associated forms. Digestive tract involvement occurs in around 40% of HIV patients [8]. KS of the digestive tract in immunocompetent subjects is rare in the literature. The specific treatment of KS is based on local means, including surgery for limited, few lesions; liquid nitrogen cryotherapy; nitrous oxide cryosurgery; fractionated-dose radiotherapy; local chemotherapy with vinblastine and general monotherapy with bleomycin. If cutaneous lesions are not extensive, treatment is local; if they are extensive and visceral lesions are not progressive, treatment is based on bleomycin monochemotherapy, 5 mg x 3 IM/14 days [2]. KS is a chronic, rather indolent condition, and its evolution is dependent on socio-economic status. The course of the disease varies from patient to patient. It is generally slow, with survival rates exceeding 20 years [9].

4. Conclusion

Non-HIV-related gastric Kaposi's sarcoma is rare in our setting. Kaposi's sarcoma of the digestive tract in immunocompetent subjects is rare in the literature. This gastric pathology should be considered even in HIV-negative patients. Surgical treatment provides lasting relief in localized forms.

Compliance with ethical standards

Disclosure of conflict of interest

No conflict of interest to be disclosed. All authors contributed to the conduct of this work. All authors also declare that they have read and approved the final version of the manuscript.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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