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Coexistent disseminated Kaposi's sarcoma and disseminated *Mycobacterium avium* complex infection in a patient with AIDS

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Late presenter;
Human herpesvirus-8;
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Immune reconstitution inflammatory syndrome

Dear editor,

Patients with AIDS are susceptible to a multitude of opportunistic illnesses, but cases of coexistent Kaposi's sarcoma (KS) and atypical mycobacterial infection in the same lesions are rarely reported.[1] We herein describe the first case of coexistent KS and *Mycobacterium avium* complex (MAC) infection in multiple lesions in an HIV-positive patient, who subsequently developed KS inflammatory cytokine syndrome (KICS).

A 33-year-old man first presented to our hospital due to abdominal pain. Five months earlier, he had received the diagnosis of AIDS with a nadir CD4 count of 18 cells/μL at another hospital and started coformulated dolutegravir, abacavir, and lamivudine. He had been treated for bone marrow infection with *M. fortuitum* complex. Due to persistent fever, a prednisone-equivalent dose of >0.4 mg/kg/day was administered for 4 months after a diagnoses of immune reconstitution inflammatory syndrome (IRIS) was made.

One month before this evaluation, purplish patches had gradually grown on the four extremities. Two days prior to admission, lower abdominal pain occurred with high fever. Physical examination was remarkable for moon face with a buffalo hump, lymphadenopathy at the left neck and inguinal regions, hepatomegaly, and tenderness at the right lower quadrant of the abdomen. Laboratory testing showed hemoglobin 6.8 g/dL, platelet 59,000 cells/μL, C-reactive protein 6.54 mg/dL, albumin 3.1 g/dL, and alkaline phosphatase 147 U/L. The low adrenocorticotropic hormone and cortisol levels suggested secondary adrenal insufficiency. His CD4 count was 18 cells/μL though plasma HIV RNA load was 57 copies/ml. Computed tomography (CT) revealed solid nodules in bilateral lungs, focal bowel wall thickening, and generalized lymphadenopathy (Fig. 1a). Colonoscopy showed a circumferential tumor with luminal stenosis at peri-ileocecal valve area (Fig. 1b). Pathological examination of biopsies from the lung nodule, colonic tumor, and lymph nodes showed spindle cells with slit-like vascular spaces and extravasated red blood cells. The immunohistochemical staining of spindle cells was positive for KS-associated herpesvirus (KSHV), and plasma KSHV DNA load was 595,000 copies/ml. Subsequently, the cultures of biopsy specimens of the lung nodule and lymph nodes, and blood, stool, sputum specimens all yielded *M. avium* complex (MAC).

According to the clinical manifestations and laboratory investigations, the patient fulfilled the working case definition of KICS.[2] The systemic inflammatory response might be attributed to both KICS and MAC infection. After administering the first dose of liposomal doxorubicin, his abdominal pain dramatically improved and fever resolved. After 4 courses of liposomal doxorubicin, the purplish patches on the extremities resolved, and the follow-up colonoscopy and CT showed significant improvement in all lesions. Antibiotics were switched to rifabutin, clarithromycin, ethambutol, and moxifloxacin for disseminated MAC infection, and steroids were slowly tapered off during hospitalization. He subsequently received outpatient follow-up with viral suppression and a CD4 count increase to 170 cells/μL 8 months later.

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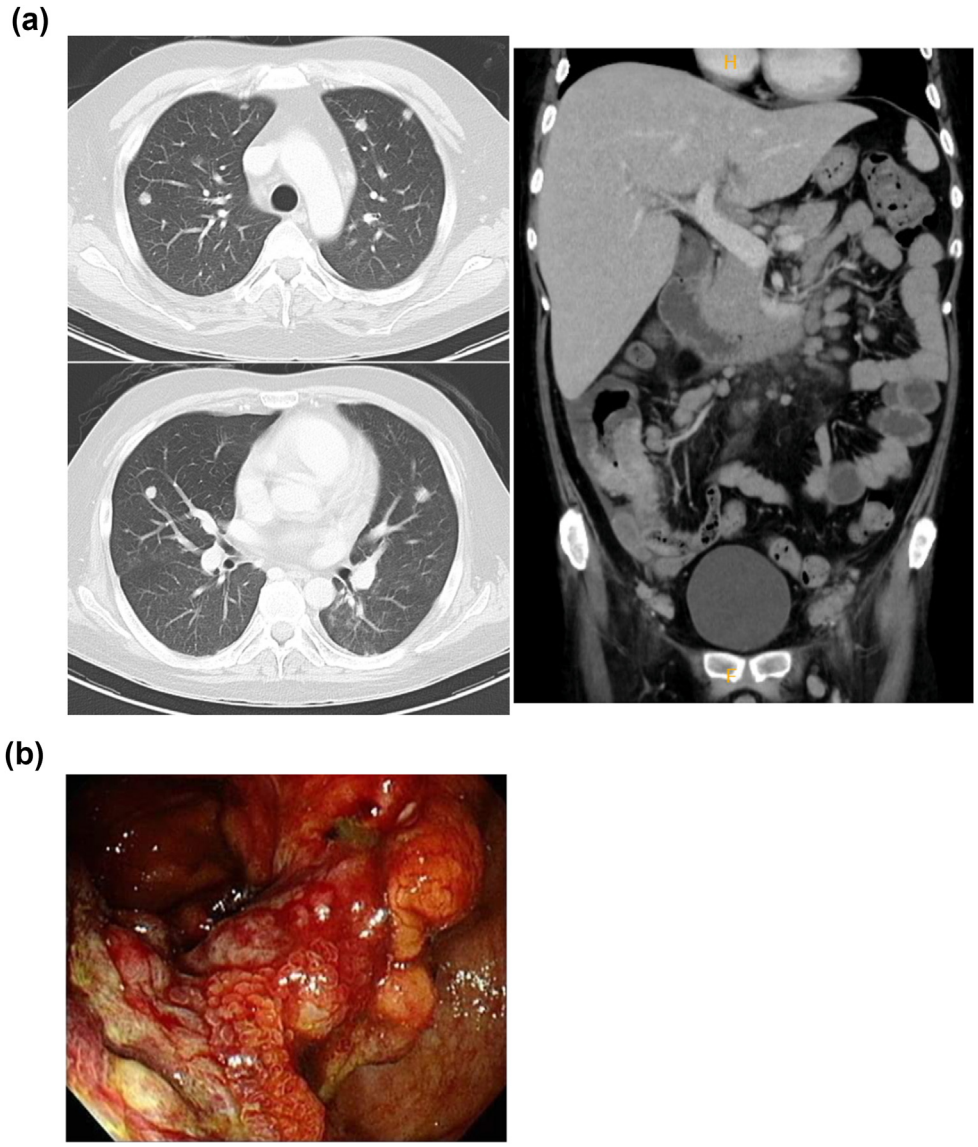

Figure 1. (a) Computed tomography of the chest, abdomen, and pelvis revealed randomly distributed solid nodules in bilateral lungs, wall thickening at the proximal ascending colon and distal ileum, as well as multiple mediastinal, hilar, abdominal, and pelvic lymphadenopathy. (b) Colonoscopy showed a circumferential tumor larger than 5 cm at peri-ileocecal valve area, with fragile mucosa and causing luminal stenosis.

Although the exact pathogenesis mechanism of coexistent infections was not well understood, the induction of KS and/or exacerbation of pre-existing KS may be associated with opportunistic infections through the release of inflammatory cytokines and oncogenic signals.[1,3] In addition, prolonged exposure to glucocorticoids may also induce reactivation of KSHV, resulting in the development of KS and KICS.[4] In our case, the differentiation between IRIS and KICS was the key to avoiding prolonged glucocorticoid use. The present case was unlikely to have IRIS considering his sustained, low CD4 counts for several months.[5] Monitoring the symptoms of KS, obtaining tissue diagnosis, and measuring KSHV DNA load facilitate early recognition and treatment of KICS.[2]

Declaration of competing interest

All authors have no potential conflict of interest.

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