ASSOCIATION OF CASTLEMAN DISEASE AND KAPOSI SARCOMA SUCCESSFULLY TREATED WITH WEEKLY PACLITAXEL

Background: Multicentric Castleman disease (MCD) or giant lymph node hyperplasia is a rare disorder first explained by Castleman et al., in 1954. It mostly involves mediastinum (60%), retroperitoneum (11%), and axilla (4%) [1]. Human herpes virus 8 (HHV-8) infection has been identified in nearly 100% of HIV-positive and 50% of HIV-negative MCD patients [2, 3]. This virus predisposes patients to much higher risk of other malignancies, including Kaposi’s sarcoma (KS) (13%) [4]. Classic Kaposi’s sarcoma (CKS) is an angioproliferative disorder that is thought to develop from endothelial cells, myofibroblasts, and monocyte-macrophages. Systemic chemotherapy is indicated for rapidly progressive CKS.

Case presentation: We report here a 71-year-old man who presented an HHV8+ Kaposi sarcoma in the background of multicentric Castleman disease which was successfully treated by weekly paclitaxel.

Conclusion: Weekly Paclitaxel is effective and tolerable as first-line chemotherapy in treating classical Kaposi sarcoma associated with multicentric Castleman disease.

Case report:
A 71-year-old man was admitted to our service with fever, anorexia, weight loss, axillary nodes, and multiple cutaneous lesions on the legs. Physical examination showed multiple violaceous plaques of varying sizes and bilateral lymphedema on his legs (fig1). The lymph nodes were discrete, mobile and firm. Human immunodeficiency virus (HIV) antibody was negative. Thoraco-abdomino-pelvic scans showed mediastinal and right axillary nodes without visceral lesions (fig2). Axillary node biopsy was performed with pathological findings of HHV8+ Kaposi sarcoma in the background of multicentric Castleman’s disease (fig3). An immunohistochemical (IHC) staining for CD34 was positive (fig4).

The patient was treated with Paclitaxel 80mg/m² weekly. There were no side effects reported. After two months of chemotherapy, we observed a major decrease in leg lesions and edema (fig5).

Discussion:
Classic Kaposi’s sarcoma is a cutaneous disease of the extremities affecting predominantly elderly men of Mediterranean origin. This form is associated with immune system alteration and malignant diseases without HIV infection [5].

Patients with extensive or recurrent KS can be treated with systemic chemotherapy. A number of drugs approved for treatment of AIDS-associated KS have activity against CKS after failure of prior therapy. These include Vinblastine, Bleomycin, Doxorubicin, and Etoposide alone or in combination [6]. Paclitaxel has also shown efficacy with minimal toxicity in patients with classical KS after failure of prior therapy [7, 8].

MCD has been reported in adults usually before age 30 [9] and shows an association with HHV-8 and HIV infections [10]. In contrast, our patient was a 71-year-old man and compared with the prior literature findings, he had HHV-8 and HIV-negative serology. Co-incidence of MCD with KS in our case report can be interpreted that MCD may be complicated by KS or it is a manifestation of MCD. MCD should be kept in mind as a differential diagnosis in a patient with KS [11].

Conclusion:
The case reported here would suggest that weekly Paclitaxel is effective as first-line chemotherapy in treating classical KS associated with MCD. The low toxicity of this regimen is well tolerated in elderly patients with comorbidities.

Consent:
Written informed consent was obtained from our patient for publication of this case report and any accompanying images.

Ethics approval:
Not applicable

Authors’ Contribution:
YS and CE participated in the literature review. AR reviewed the manuscript and revised it critically for important intellectual content. AD carried out the literature review. YS and CE participated in the literature review. AR performed pathological examinations. HE and MI carried out the conception of the case, revised it critically for important intellectual content. All authors read and approved the final manuscript.

Competing interests:
The authors declare that they have no competing interests.

Funding Section:
We have no source of funding to declare.

Availability of data and supporting materials:
Medical imaging data will not be shared because it is not fully anonymous.

ABSTRACT

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Background: Co-existence of multicentric Castleman disease and classic Kaposi sarcoma has been rarely reported in medical literature, especially if HIV is negative.

Case presentation: We report here a 71-year-old man who presented an HHV8+ Kaposi sarcoma in the background of multicentric Castleman disease which was successfully treated by weekly paclitaxel.

Conclusion: Weekly Paclitaxel is effective and tolerable as first-line chemotherapy in treating classical Kaposi sarcoma associated with multicentric Castleman disease.

KEY WORDS: Homomorphism token, Distributed scheme, Data redundancy, cloud.
Fig 1: Violaceous indurated classic Kaposi's sarcoma lesions on the legs

Fig 2: Computed tomography of the thorax revealed right axillary nodes.

Fig 3: HHV-8 antibody: nuclear positivity of spindle cells

Fig 4: Positivity of CD34 on the spindle cells of vascular tumor proliferation

Fig 5: Evolution of the lesions after treatment by Paclitaxel.

References