Unusual Involvement of Postrenal Transplantation Kaposi's Sarcoma: Two Case Reports

Introduction

Malignancy and infectious diseases are the most serious problems faced by solid-organ recipients in the long term. The average incidence of post-transplant malignancy is around 4%, and the risk of certain types of tumors is 500 times higher than that in the normal population.[1] Kaposi's sarcoma (KS) is a multicentric vascular or lymphatic endothelial cell neoplasm and predominantly occurs in immunodeficient patients who receive immunosuppressive therapy and in patients with acquired immunodeficiency syndrome (AIDS). Although the most common site of KS is skin, visceral involvement can occur less commonly. Additionally, oral KS and involvement of the transplanted organ as the initial manifestation of KS are rarely described.

Case 1

A 46-year-old man who had been on hemodialysis for 9 years had received a kidney transplant from a living donor 3 years ago. His pre-transplant serology was negative for Hepatitis B, venereal disease research laboratory (VDRL), and HHV8. He received anti-thymocyte globulin as induction therapy and tacrolimus (Tac), mycophenolate mofetil (MMF), and prednisolone (Pred) as maintenance treatment. His post-transplant follow-up was performed by his local nephrologist.

Three years after transplantation, the patient was admitted to our clinic with complaints of swelling in the legs and high serum creatinine level. During the physical examination, a nonpigmented and mucosa-colored nodular lesion on the tongue and pitting edema of the legs were noticed [Figure 1]. Neck MRI demonstrated a heterogeneously enhancing mass 5 cm in size, with a nodular component that compressed the epiglottis in the root of the tongue with two pathological appearing submandibular lymph nodes. Histopathological examination revealed squamous epithelial proliferation, keratinization, and typical regenerative hyperplasia. Immunohistochemical staining for CD34, HHV-8, and cytokeratin AE1/AE3 was positive in the tumor cells. The patient was diagnosed with posttransplant Kaposi sarcoma (PTKS) on the basis of these findings. Tacrolimus was withdrawn, Everolimus was initiated, and 50% dose reduction was made in MMF. Liposomal doxorubicin chemotherapy was planned.

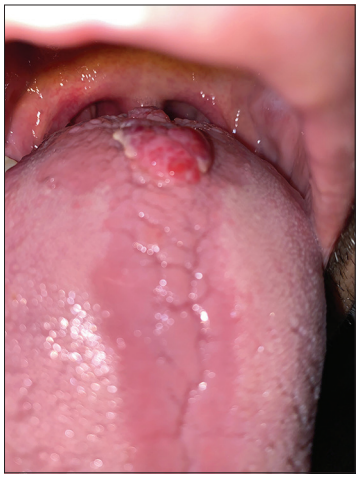

Figure 1: Nonpigmented and mucosa-colored nodular lesion on the tongue

Case 2

Another 26-year-old man who was admitted to our hospital with fatigue, lower abdominal pain, and abdominal swelling 3 months after a living-donor kidney transplant from his mother. His pre-transplant serology was negative for Hepatitis B and C, HIV, CMV, EBV, VDRL, and HHV8. The patient was discharged with triple immunosuppressive therapy with Tac, MMF, and Pred. When he was readmitted, his physical examination revealed a palpable mass displacing the transplanted kidney and palpable fixed inguinal lymph nodes.

Transplant renal biopsy and pelvic MRI were performed. While biopsies obtained from the transplanted kidney demonstrated spindle cell mesenchymal tumor with undetermined malignant potential, pelvic MRI disclosed an infiltrating tumor in the transplanted kidney, para-aortic lymph nodes, and conglomerated lymph nodes in the right obturator group with multiple metastatic bone lesions in lumbar vertebra, pelvis, and proximal femur. PET-CT scan showed increased FDG uptake on transplanted kidney and supra and infra-diaphragmatic nodes; moreover, bone lesions were defined on MRI. For definitive diagnosis, an excisional inguinal lymph node biopsy was performed and metastasis of Kaposi sarcoma was detected. MMF was withdrawn, and the patient was started on Everolimus and liposomal doxorubicin chemotherapy.

Discussion

KS is an HHV-8-related vascular disease that is seldom seen in immunocompetent adults and generally presents with skin lesions such as pink-purple nodules and plaques on legs, arms, and trunk. Although skin involvement is found in 90% of patients diagnosed with KS, extracutaneous sites, including lymph nodes, gastrointestinal tract (most commonly stomach and duodenum), spleen, and lung, are described in 10% of recipients.[2] In the literature, oral

mucosa involvement was reported previously in 14 patients in the non-AIDS-associated KS group.[3] Two patients who received kidney transplants from HHV-8 positive donors and were diagnosed with KS during post-transplantation follow-up had been reported previously.[4] This case demonstrates that KS may develop at any organ site. Oral KS, which was manifested with a purple lesion on the tongue and hard palate, was previously reported in a HIV-positive patient.[5] However, recognition is difficult when nonpigmented and mucosa-colored KS lesions are limited to the oral cavity.

Although PTKS has been described in patients after receiving a kidney from a HHV8 seropositive donor, the donor tested negative for HHV-8 in our case. In line with transplantation literature, PTKS appeared 3 months after transplantation in our second case, whereas it occurred 3 years after transplantation in our first case, which is quite a long time for PTKS development. To our knowledge, primary occurrence of PTKS in tongue and in the organ transplanted from HHV-8 seronegative donor has not been previously described in Turkish literature.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Statement of ethics

An informed consent from the patients were obtained.

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Conflicts of interest

There are no conflicts of interest.

Author Contributions

All authors contributed equally to the study and writing of the paper.

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