Solitary Laryngeal Kaposi Sarcoma in a Kidney Transplant Patient

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After the first description of Kaposi sarcoma in 1872, many cases of this tumor were reported worldwide. This tumor is multifocal and laryngeal involvement is considered to be as unusual site. Kaposi sarcoma is almost always are associated with classical skin lesion, and only about 5% of non-acquired immune deficiency syndrome Kaposi sarcomas are reported to be located in the larynx. We report a kidney transplant recipient diagnosed with solitary laryngeal Kaposi sarcoma 21 months after transplantation, who was treated with combined surgery, chemotherapy, and immunosuppressive modification.

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Case Report

INTRODUCTION

Kaposi sarcoma is an angiogenic tumor characterized by endothelium-lined vascular spaces and spindle-shaped cells and is usually seen in immunocompromized conditions such as acquired immune deficiency syndrome (AIDS) or organ transplantation. In this report, we present a kidney transplant patient with Kaposi sarcoma in an unusual site.

CASE REPORT

A 40-year-old man presented with severe hoarseness for a month. He had received a kidney transplant 21 months earlier. On physical examination, no remarkable finding was noted, except for mild sore throat. He had developed chronic glomerulonephritis, treated with prednisolone, and chronic kidney disease from 1989 till 2005 when he started hemodialysis. In his pretransplant evaluation, viral markers for hepatitis B, hepatitis C, human immunodeficiency virus, cytomegalovirus, and Epstein-Barr virus were negative, except for immunoglobulin M antibody for Epstein-Barr virus and cytomegalovirus. After transplantation, for about 1.5 months, the patient had increased blood glucose level which was treated with insulin. With decreasing prednisolone and cyclosporine doses, his blood glucose was normalized and insulin was discontinued.

The patient was referred to the otolaryngology clinic, where after routine examination and was treated with antihistamine and antibiotics. Two months later, the patient returned with worsening of the symptoms and was again was referred to the otolaryngology clinic. Direct laryngoscopy revealed a mass on the vocal cord. On contrast spiral computed tomography of the neck (Figure 1), a 20 × 18-mm polypoid mass was seen in the left side of the glottic space with invasion to the anterior commissure. True and false vocal folds were also involved. Biopsy of the lesion was performed. Sections of laryngeal tissue showed squamous epithelium and underlying struma. Neoplastic mesanchymal cell proliferation with fascicular pattern and vascular slit-like spaces and channels filled with erythrocytes were seen. Pleomorphism, atypism, and mitosis were detected in small amounts (Figure 2). On immunohistochemical staining, CD34 and vimentine were positive and actin and CK were positive (Figure 3).

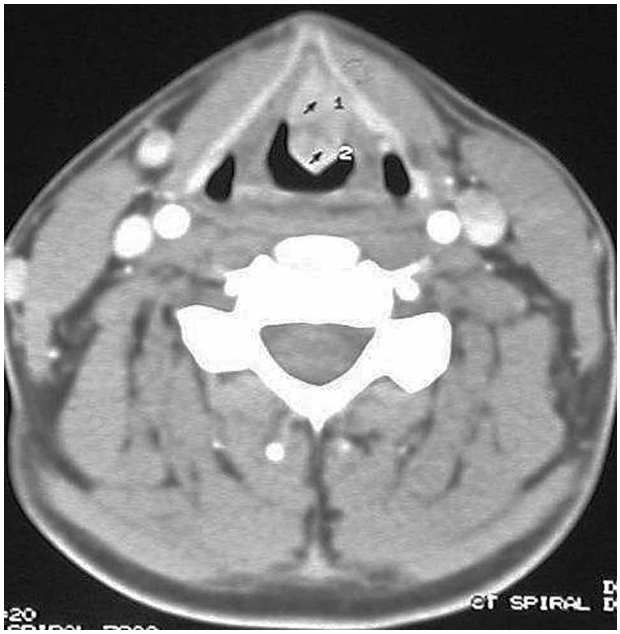

Figure 1. A polypoid mass (arrows) in the left side of the glottic space with invasion to anterior commissure.

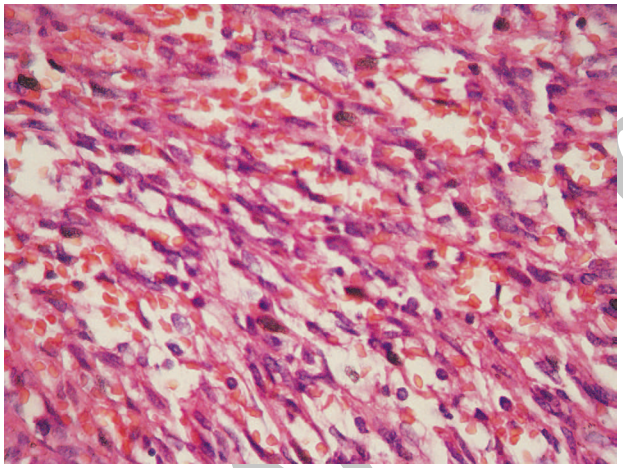

Figure 2. Microscopic appearance of Kaposi sarcoma. Elongated spindle cells showing minimal atypia are separated by slits containing erythrocytes (hematoxylin-eosin, × 400).

A diagnosis of Kaposi sarcoma was established. The lesion was excised through transoral laser operation. Chemotherapy protocol included dexamethason and paclitaxel. Immunosuppressive regimen was changed from cyclosporine, mycophenolate mofetil, and prednisolone to sirolimus, 2 mg/d, and prednisolone, 5 mg/d. The patient's condition completely improved, and after about 3 years of close medical follow-up, no recurrence or metastasis was detected. After switching to sirolimus, his blood glucose level increased, needing treatment with metformin and

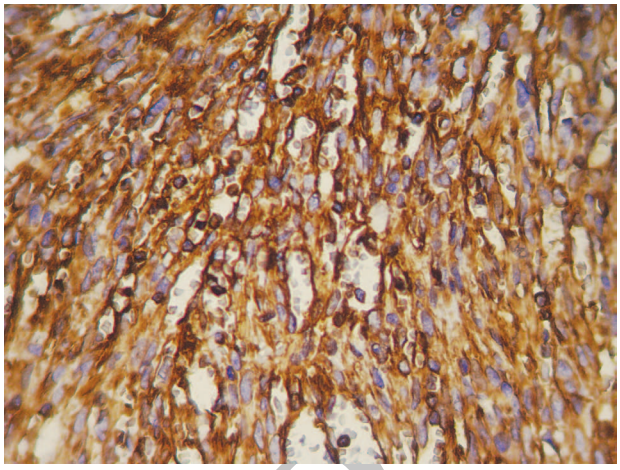

Figure 3. Immunoreactivity for vimentin in neoplastic spindle cells and vascular endothelial lining.

moderate lower extremity edema managed with losartan and furosemide. His last serum creatinine was 1.3 mg/dL, and proteinuria (268 mg/24 hours) was detected.

DISCUSSION

After the first description of Kaposi sarcoma by Moritz Kaposi in 1872, many cases of this tumor were reported worldwide.[1] Kaposi sarcoma is classified into 4 types based upon the clinical circumstances in which it develops: classic, endemic, immunosuppression associated or transplantation associated, and epidemic or AIDS associated.[2] This tumor is multifocal that manifests most frequently in mucocutaneous sites, oropharyngeal mucosa, lymph nodes, and visceral organs, most notably the respiratory and gastrointestinal tracts. Laryngeal involvement is considered to be as unusual site.[3] Almost all reported cases of this location are in patients suffering from advanced AIDS.[3-7] In a review by Patrikidou and colleagues, only about 5% of non-AIDS Kaposi sarcomas were located in the larynx.[2] Liao and coworkers reported a kidney transplant recipient with solitary laryngeal Kaposi sarcoma, who was managed with surgery and immunosuppressant reduction.[8] In our report, a kidney transplant recipient, 21 months after transplantation diagnosed with solitary laryngeal Kaposi sarcoma and treated with combined surgery, chemotherapy, and discontinuing cyclosporine and mycophenolate mofetil replaced with sirolimus, resulting in complete remission of the tumor without recurrence after 3 years of follow-up. Kidney function was preserved but emergence of

diabetes mellitus had complicated the situation. In conclusion, Kaposi sarcoma is a well-known complication of kidney transplantation, but unusual sites like larynx should not be forgotten.

CONFLICT OF INTEREST

None declared.

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