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Case report

A case of post-transplant lymphoproliferative disorders that developed ten years after living renal transplantation in the parotid gland region

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ABSTRACT

Post-transplant lymphoproliferative disorders (PTLDs) are lesions that proliferate lymphocytes or plasma cells and are caused by immunosuppression after organ transplantation or hepatopoietic stem cell transplantation. According to the 2016 WHO classification, PTLDs are classified into four types: early lesions, polymorphic PTLDs, monomorphic PTLDs, and classical Hodgkin lymphoma-type PTLDs. We report a case of PTLD that first occurred in the oral and maxillofacial area more than ten years after living renal transplantation. A sixty-one-year-old man visited our department in the middle of August 2013 because of right parotid gland swelling. We consulted with an otorhinolaryngologist and prescribed antibiotics to determine whether the swelling was parotitis or a tumor; however, the swelling did not improve. At the end of September 2013, a tumor was found in the right palatine tonsil; consequently, a biopsy was performed. After the biopsy, the lesion was diagnosed as monomorphic PTLD (diffuse large B-cell lymphoma), and the patient was admitted to the hematology and oncology ward. Immunosuppressants were interrupted or reduced. However, the tumor continued to swell, and R-CHOP treatment was started. The therapeutic value was PR, and the patient's general condition was good; therefore, in mid-February, he discharged. However, at the end of April 2014, the patient experienced septic shock due to pneumonia and passed away the next day. As this case shows, it is important for oral and maxillofacial surgeons to consider the possibility of PTLD in the oral and maxillofacial area.

Introduction

Post-transplant lymphoproliferative disorders (PTLDs) are lesions that proliferate lymphocytes or plasma cells and are caused by immunosuppression after organ transplantation or hepatopoietic stem cell transplantation. According to the 2016 WHO classification, PTLDs are classified into four types: early lesions, polymorphic PTLDs, monomorphic PTLDs, and classical Hodgkin lymphoma-type PTLDs. However, small B cell tumors and MALT lymphoma are not included among PTLDs, even if they develop after transplantation [1]. Although the 5-year cumulative incidence rate of PTLDs in living body renal transplant patients is comparatively low, ranging from 1 to 3%, many cases develop within 1 year after transplantation due to the use of numerous immunosuppressants [2].

In this writing, I report a case of PTLD initially discovered in the maxillofacial area more than 10 years after the transplantation of a kidney from a living body.

Case report

A sixty-one-year-old man visited our department in the middle of August 2013 because of right parotid gland swelling. He had experienced some prior medical problems. In 2003, living transplantation was performed at our hospital because of chronic renal failure. Since then, the patient had taken Neoral® 130 mg/day, prednisolone® 5 mg/day, and Sercept® 1000 mg/day. Additionally, he presented amyloid hip arthropathy resulting from long-term dialysis. In 2010, he began continuous nasal positive pressure respiration treatment for obstructive sleep apnea syndrome (OSAS). In 2012, he underwent posterior cervical spinal fusion surgery to remedy cervical spondylosis.

During the first examination, swelling of elastic hardness and poor mobility of 30 × 30 mm in the right parotid region and 20 × 20 mm in the right cervical region was observed. B symptoms were not observed. No obvious organic change was observed in his oral cavity. In particular, no redness or swelling was found in the buccal mucosa around the papilla of the right parotid gland. A 38 × 36 mm area of radiopacity with marginal irregularities was observed in the medial portion of the

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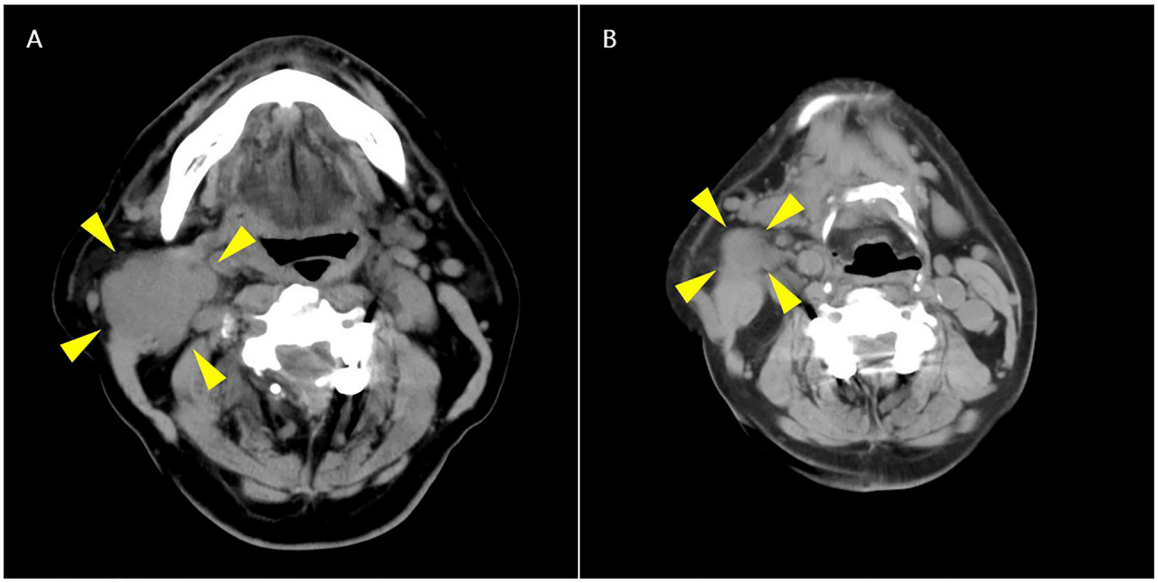

Fig. 1. Computed tomography imaging.
A : A 38 × 36-mm tumor was found inside the right parotid gland.
B : A 22 × 20-mm tumor was found in the right superior internal jugular lymph node.

right parotid gland on the CT image; borders on the right upper mediastinal jugular vein lymph node were bounded clearly with 22 × 20-mm margin alignment (Fig. 1A, B). Ultrasound images revealed a nonuniform tumor of 25 mm in size with marginal irregularities inside in the right parotid gland and a 20-mm tumor with a nonuniform interior and margin alignment in the right cervical region. Laboratory data showed some abnormal values (LDH: 264 IU/L. WBC: 8600 / μl, CRP: 0.52 mg/dl). Other data were within normal limits. Given these findings, we suspected a right parotid lesion or malignant lymphoma.

In mid-August 2013, we consulted with the department of otorhinolaryngology because of the possibility of parotid tumor, and we prescribed antibiotics to rule out either parotid inflammation or neoplastic lesions. By late August, the swelling had not improved; therefore, a tumor was suspected. Fine needle aspiration (FNA) was then performed; however, a clear diagnosis was not obtained. In early September, magnetic resonance imaging (MRI) was performed; the lesion in the right parotid region had grown to a size of 45 × 42 mm, a clear increase compared with the previous CT image. However, the lesion was in the deep part of the parotid region, making it difficult to perform biopsy. We opted to perform FNA again; however, it was not possible to obtain a clear diagnosis, although PTLD was suspected. In late September, we found a tumorous lesion in the right palatine tonsil (Fig. 2), so we performed biopsy under local anesthesia. Pathological examination yielded a diagnosis of monomorphic PTLD (diffuse large B cell lymphoma; Fig. 3,4). The patient was admitted to the department of hematology. FDG-PET images taken the day before hospitalization showed abnormal accumulation from the right parotid gland to the tonsils and neck, and SUVmax was 37.2. Although no abnormal accumulation was found in other organs, the maximum diameter of the tumor was 76 mm, a large increase over the previous condition. Although the patient had been taking some immunosuppressants, prescriptions for Neoral® and Sercept® were discontinued, and the patient's intake of prednisolone® was reduced. Given the rapid growth of the tumor, R-CHOP therapy was started. In February 2014, the therapeutic value was PR (Fig. 5). The patient was discharged at the end of February because his general condition was good. However, he developed a fever and new symptoms, including hallucinations, in late April of the same year and was brought to our hospital by ambulance. He was diagnosed with septic shock caused by pneumonia and was treated accordingly, but unfortunately, he passed away the next day.

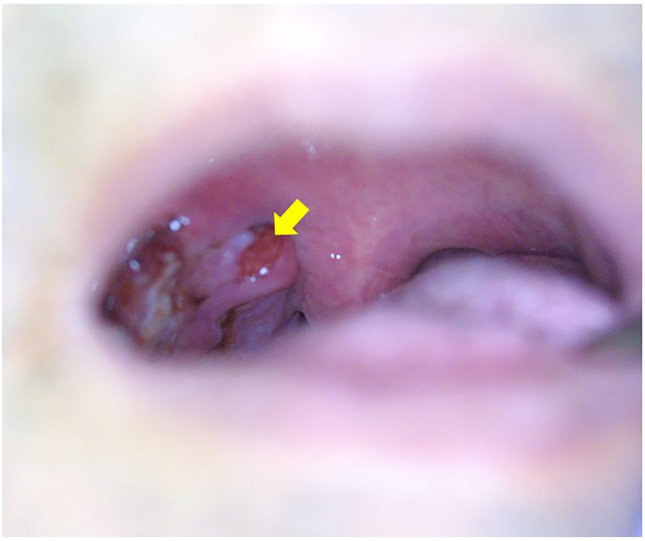

Fig. 2. A tumor was found in the right palatine tonsil.

Discussion

More than 1000 kidney transplantations are performed in our country [3]. Transplantation is among the most useful treatment methods for many end-stage renal failure patients, including hemodialysis patients. However, a number of patients have reported malignant tumors as an adverse event after kidney transplantation [4]. According to Farrugia et al. [5], malignant disease accounted for approximately 18% of the deaths of 19,103 renal transplant recipients. In addition, it is said that the carcinogenesis rate of renal transplant recipients is 15 to 30 times that of healthy people. Immunological monitoring mechanisms suggest that the increase in the proportion of patients suffering from malignant diseases is linked to the existence of virus-related tumors caused by immunosuppressants used after surgery and the suppression of sensitized lymphocytes against tumors [6].

The risk factors include (1) serologic inconsistencies in EBV and CMV among recipients and donors, (2) transplantation during childhood, (3) HLA incompatibility, (4) anti-T cell therapies with OKT3, and