Primary omental pleomorphic liposarcoma identified at autopsy in a living donor kidney transplant recipient

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Abstract Metastatic and recurrent tumors of the omentum are common, but primary omental pleomorphic liposarcoma (POPL) is an extremely rare type of solid omental tumor. We describe the case of a patient with POPL who received a renal transplant from a living donor. Despite good allograft function, the volume of peritoneal fluid gradually increased. An exploratory laparotomy could not be performed because the patient was obese and developed hemodynamic instability. Therefore, a shunt was placed between the peritoneal cavity and the internal jugular vein using the Denver® shunt system; however, the patient died of respiratory insufficiency. On the basis of the autopsy results, we diagnosed the patient's condition as POPL. We speculated that the malignancy did not originate directly from the donor cells. We report POPL in a living donor renal transplant recipient.

Keywords Primary omental tumors · Pleomorphic liposarcoma · Living renal transplantation · Continuous ambulatory peritoneal dialysis

Introduction

The incidence of malignancy after transplantation is high and is a major problem associated with the administration

of immunosuppressive therapy [1]. Metastatic and recurrent tumors of the omentum are common, but primary tumors of the omentum are relatively rare [2]. Primary tumors of the omentum were first described by Stout in 1943 as a hemangiopericytoma [3]. Liposarcoma has been reported in only a few patients receiving maintenance hemodialysis [4, 5], and the worldwide occurrence of liposarcoma is unknown. To our knowledge, this is the first report on primary omental tumor after renal transplantation.

We report the case of a patient who underwent living donor renal transplantation with primary omental pleomorphic liposarcoma (POPL) identified at autopsy.

Case report

A 63-year-old Japanese man was followed up after living donor renal transplantation from his wife in October 2006. Renal graft function was well maintained (serum creatine level 1.6–1.8 mg/dL). He was treated with continuous ambulatory peritoneal dialysis (CAPD) for diabetic nephropathy from 2003 until renal transplantation in 2006. Because of continued slight fever and abdominal discomfort beginning in October 2010, he was admitted to our center for the examination of an acute inflammatory reaction and increased ascitic fluid levels in November 2010.

On admission, the patient was 171 cm tall, weighed 101.7 kg, and had a body mass index of 34.8. Computed tomography showed thickened omentum, a large volume of ascitic fluid, and no evidence of a mass in the peritoneal space (Fig. 1). The levels of tumor markers were within normal limits (carcinoembryonic antigen, 2.4 ng/mL; α -fetoprotein, 3.0 ng/mL, and carbohydrate antigen 19-9, 6.0 ng/mL). Examination of the peritoneal fluid several

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Fig. 1 Computed tomography demonstrating thickening omentum, a large volume of ascites, and no clear mass in the peritoneal space (**a, b**). The transplanted kidney allograft was in the right iliac fossa (**c**)

times showed the following values: cell count 600 cells/ μ L (neutrophils:lymphocytes 400:200) and adenosine deaminase 9.9 IU/L; cultures for bacteria and acid-fast bacterium were negative, and cytology was consistently class IIIa. Initial treatment using antibiotics was ineffective and, thus, steroids and gamma globulin were included. Steroids temporarily lowered the ascetic fluid volume. We used a hemodiafiltration system three times weekly after 3 weeks of hospitalization because the amount of peritoneal fluid gradually increased and renal function worsened. Additionally, we placed a shunt between the peritoneal cavity and the right internal jugular vein using the Denver[®] shunt system to enhance hemodynamic stability so that hemodiafiltration could be performed effectively. The ascites pump was efficient and stable, but the patient died of respiratory insufficiency because of lung congestion and pneumonia after 10 weeks of hospitalization.

Pathological findings

The patient's abdominal girth was 130 cm and the abdomen contained 11.7 L of peritoneal fluid. Histopathological examination revealed that the omentum was covered with small nodules, thickened at the intestine (Fig. 2), and had intensely proliferated malignant pleomorphic cells that had progressed to partial necrosis (Fig. 3). Immunostaining showed that the cells were positive for vimentin and some were also positive for S-100 (Fig. 4). The tumor had

metastasized to the liver, gallbladder, small intestine, pleura, and lymph nodes.

Discussion

In organ transplant recipients, malignancies are the most common cause of mortality. The overall occurrence of malignancies after organ transplantation is approximately 10 %, and transplant recipients have a three- to five-fold higher risk of malignancies than the general population [1]. Skin tumors are common in transplant recipients from Western countries, but the types of malignant disease differ. In Asian patients, gastrointestinal tumors are the most common type of malignancies [1], while the incidence of liposarcoma is fairly rare. In transplant recipients, the determination of whether the tumor cells are derived from the donor cells is important.

Metastatic and recurrent tumors of the omentum are common, whereas primary tumors of the omentum are rare [2]. Omental tumors are commonly observed in leiomyosarcoma, hemangiopericytoma, and fibrosarcoma, but liposarcoma occurs in only 1 % of patients [6]. Some studies report that primary omental tumors are most common in men aged 50–60 years [7, 8]. Primary liposarcoma of the omentum is not associated with any specific symptoms and, therefore, diagnosis and management is often delayed. Liposarcomas appear as poorly demarcated infiltrative masses containing fat and soft tissue components on

Fig. 2 Omental liposarcoma. Resected specimen at autopsy of the patient. Omentum was covered with small nodules and developed thickening upon the intestine (a). H&E staining of the omentum, low-power view of pleomorphic malignant cells with necrotic areas (b)

Fig. 3 High-power view of the malignant lipoblasts (a, b). Silver (c) and oil red O (d) staining of the lipoblasts

Fig. 4 Immunohistochemical staining of pleomorphic sarcoma, showing positive reaction for vimentin (a) and some cells positive for S-100 (b)

computed tomography and magnetic resonance imaging, and malignant tumors are more than 5 cm in diameter, deep-seated, firm, and fixed [9, 10]. According to the 2002 World Health Organization (WHO) classification,

liposarcomas are categorized into five groups: myxoid-cell, round-cell, well-differentiated, dedifferentiated, and pleomorphic type. The pleomorphic type is the rarest type, occurring in only 5–10 % of liposarcoma patients.

Furthermore, pleomorphic liposarcoma has clinical features of high-grade tumors, with a high risk of local recurrence and metastasis [11, 12].

In the present case, we considered different potential causes of the malignant disease, such as infection and encapsulating peritoneal sclerosis due to CAPD, but we were unable to identify the reason for the intractable ascites. Computed tomography did not reveal a clear mass in the peritoneal space, the patient was negative for tumor markers, and cytology was consistently class IIIa. Therefore, the diagnosis and selection of the appropriate treatment option at an early stage of the disease was difficult. Because the patient was obese and developed hemodynamic instability, an exploratory laparotomy could not be performed. Surgical treatment involved a high risk and, therefore, the tumor was managed using a peritoneal-venous shunt. Resistance to steroids and the administration of immunosuppressive agents after renal transplantation increase the incidence of development of malignancies. Thus, diagnosis at an early stage is imperative for performing surgery. POPL has a less favorable prognosis and is associated with treatment difficulties encountered after the operation. Additionally, we could not investigate the chromosome expression. More than 4 years have passed since the patient has undergone renal transplantation, and the tumor was located only in the peritoneal cavity and did not extend around the allograft into the retroperitoneal space. Therefore, we speculated that the malignant tumor did not originate directly from the donor cells.

Conflict of interest The authors of this manuscript have no conflicts of interest to disclose.

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