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Leiomyosarcoma after Kidney Transplantation in a Patient with Leukocytoclastic Vasculitis, Lobular Panniculitis and History of Hyperthyroidism

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Abstract Leiomyosarcoma is a rare, aggressive malignant connective tissue cancer of mature adults, which increases from smooth muscle and occurs most frequently in the uterus, bowel, vascular tissues, and less commonly in somatic soft tissue or bone. We reported a 35-year-old woman was referred to the gastroenterology service with abdominal pain and vomiting. She was a case of renal transplantation in right side since 5 years ago. An evaluation by ultrasound showed right chest wall soft tissue mass measuring 40x36mm with internal vascularity without rib destruction. The CT Thorax without contrast, showed a solid mass in the right lower thoracic wall measuring 42x32mm with invasion to subcutaneous fat and abdominal wall muscle. The patient had history of hyperthyroidism about two years ago, which was treated and history of cutaneous leukocytoclastic vasculitis (LCV) and lobular panniculitis since last year, which was proven by skin biopsy. Our patient sought surgical advice who asked for a biopsy which showed a malignant spindle cell tumor compatible with leiomyosarcoma extending to peripheral fatty tissue with histological grade 2. In conclusion, treatment of thyroid disorders and LCV in kidney transplanted patients can be caused malignancies. Therefore, evaluation for malignancies like sarcoma is suggested with thyroid disorders and vasculitis in the follow-up of kidney transplanted patient.

Keywords: *leiomyosarcoma, leukocytoclastic vasculitis, kidney transplantation*

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1. Introduction

Smooth muscle tumors including leiomyosarcoma, in contrast to malignant lymphomas or skin cancer, are rarely associated with transplant recipients [1]. Sarcomas of soft tissues are rare tumors accounting for approximately 1% of all adult malignancies and account for a 1.7% incidence among all transplanted patients presenting with de novo malignancies [2]. Sarcomas in solid organ transplantation appear to have an aggressive pattern, with 62% being high grade and 40% metastatic at the time of a primary diagnosis [1]. A number of studies found that the most common malignancy after kidney transplantation was Kaposi Sarcoma among the Iranian patients [3]. Leiomyosarcoma is a rare, aggressive malignant connective tissue tumor of mature adults, which arises from smooth muscle and occurs most frequently in the uterus, bowel, vascular tissues, and less commonly in somatic soft tissue or bone [4]. Vasculitis is a syndrome which may complicate certain infectious, rheumatologic, and allergic diseases [5]. Probably there is a close temporal relationship sarcoma and vasculitis [6,7]. The aim of study is to report leiomyosarcoma after kidney transplantation in a patient with leukocytoclastic vasculitis(LCV), lobular panniculitis and history of hyperthyroidism in Western Iran.

2. Case Report

A 35-year-old woman was referred to the gastroenterology service on February 17, 2016 for abdominal pain and vomiting. The pain radiated to chest and back. She underwent a renal transplantation in right side since 5 years ago. Endoscopy report was normal except for retention of food in stomach more than 12 hours after fasting which suggested gastroparesis. An evaluation by ultrasound on February 27, 2016 showed right chest wall soft tissue mass measuring 40x36mm with internal vascularity without rib destruction. Multiple masses in the left abdominal wall near umbilicus and subcutaneous fatty tissue of buttock and lateral of the thighs were also detected. The CT Thorax without contrast, showed a solid mass in the right lower thoracic wall measuring 42x32mm with invasion to subcutaneous fat and abdominal wall muscle. In deep subcutaneous fat of left anterior costal margin smaller solid mass with

attachment to adjacent muscular wall with no rib destruction or deep extension also was noted.

The patient underwent CT Abdomen and pelvis with contrast and abdominal CT-angiography with and without contrast which showed a solid 42mm mass in abdominal wall adjacent to anterior lower ribs and similar mass measuring 28mm in the lower portion and lateral to the umbilicus in the abdominal wall. Many smaller masses in the subcutaneous fat of the abdomen and pelvis were also detected. In past medical history, the patient had proteinuria and hypertension since 14 years ago, culminating to renal failure 10 years ago. She was renal transplanted 5 years ago after 6 months of hemodialysis. The patient had history of hyperthyroidism about two years ago, which was treated and history of cutaneous LCV and lobular panniculitis since last year, which was proven by skin biopsy (Figure 1). In family history, there was hypertension and hyperlipidemia in her father. In drug history after transplantation the patient had received cyclosporine-A 125mg BID, cellcept 1000mg BID and prednisolone 15mg daily. Other drugs were calcitriol, folate, amlodipine, acyclovir and nitrofurantoin. In the last admission on February 17, 2016 drugs were Cyclosporine-A,Prednisolone, Tacrolimus, Hydroxychloroquine, Pantozol, Folate, Calcium and Cellcept. Our patient sought surgical advice who asked for a biopsy. The surgery of right side chest wall mass was done on March 7, 2016. The specimen that was sent to pathology department consists of a tan /brown spherical mass measuring 5x4x4cm. The pathology report was "Malignant spindle cell tumor compatible with leiomyosarcoma extending to peripheral fatty tissue" with histological grade:2, mitotic score:3, necrosis:0, (total tumor score:2). Immunohistochemistry was done on paraffin blocks and showed positivity for SMA and CD99 (Figure 2) with negativity for NSE, CD34, CD68, Desmin, Bcl-2, EMA, CK and S100. The patient referred to the oncologist.

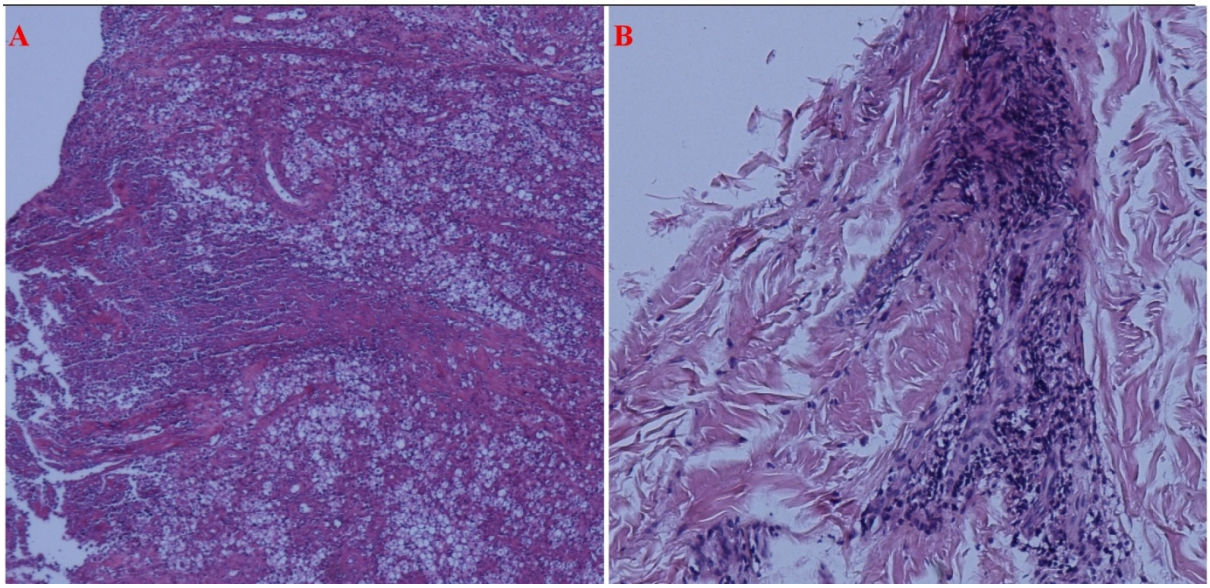

Figure 1. (A): Lobular panniculitis, skin biopsy [x40 magnification, H&E staining] (B): Leukocytoclastic vasculitis, skin biopsy [x100 magnification, H&E staining]

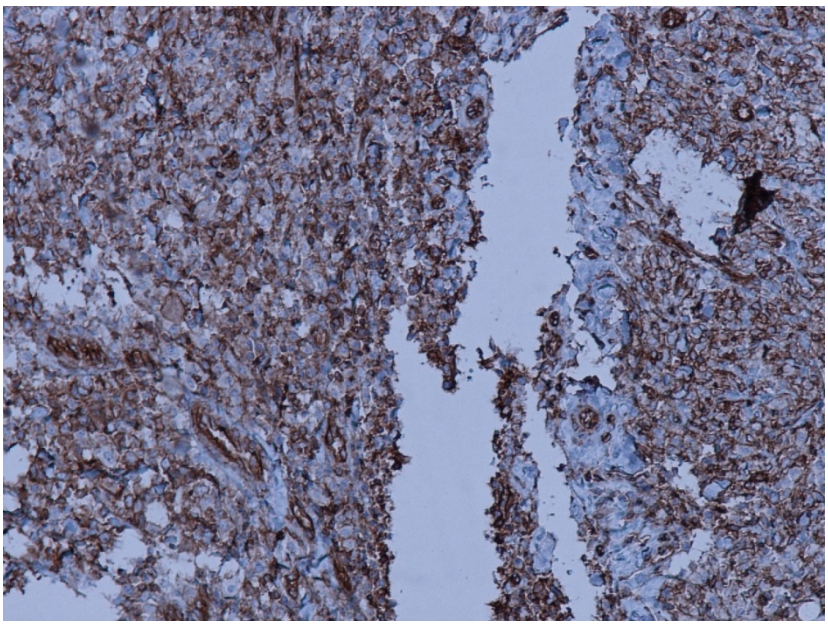

Figure 2. CD99 positivity in leiomyosarcoma [x100 magnification, Immunohistochemical staining]

3. Discussion

There are many mechanisms explaining the link between thyroid (hyperthyroidism) and kidney disease [8]. In 2010, Jeribi et al. [9] reported 22 cases of EBV-induced leiomyosarcomas after renal transplantation. According to the literature, the diagnosis of leiomyosarcoma occurs within 1–5 years after transplantation. The clinical signs depend mainly on the tumor site. One study of sarcoma patients [10], showed that many of sarcoma patients presented had thyroid disorders (goiter, thyroiditis and carcinoma). The interval between the diagnosis of the thyroid disorder and the sarcoma varied. The base of the treatment in leiomyosarcomas is surgery [11]. After that, chemotherapy such as doxorubicin, ifosfamid, gemcitabine and docetaxel and carboplatin and combination therapy of these drugs are other treatments [12]. Among renal transplant recipients, 20% of sarcomas arose at the site of an arteriovenous fistula and leiomyosarcoma in posttransplant also linked with immunosuppression [13] and EBV infection [14]. Leukocytoclastic vasculitis (LCV) is one of the most common severe side effect after antithyroid drugs consumption in patients with hyperthyroidism [15,16]. Overall, malignancy is found in approximately 5% of patients who present with vasculitis. LCV is reported as a paraneoplastic event primarily in association with hematologic malignancies. Rarely, LCV associates with solid tumors, including sarcomas [6,17]. The rarity of these associations, and in many reports the lack of temporal relationships, has led to skepticism about vasculitis being a paraneoplastic syndrome. During the 18.5 years of a study in one clinic, 0.00046% patients had both malignancy and systemic vasculitis that the most common vasculitis was cutaneous LCV [6]. Greer et al. [5] identified 13 patients, over the past 17 years, who had both vasculitis and lympho- or myeloproliferative disorders. Ten of 13 patients presented with cutaneous vasculitis antedating malignancy by an average of 10 months. Bouattar et al. [7] reported a 50-year-old woman who presented with Kaposi's sarcoma 18 weeks after starting immunosuppressive therapy for vasculitis. Also, Echeverría et al. [18] reported one patient that developed panniculitis after kidney transplantation. In this study, the patient had a treatment course for hyperthyroidism, LCV and lobular panniculitis after around 3-4 years of kidney transplantation.

4. Conclusion

Treatment of thyroid disorders and LCV in kidney transplanted patients can be caused malignancies. Therefore, evaluation for malignancies like sarcoma is suggested with thyroid disorders and vasculitis in the follow-up of kidney transplanted patient.

Competing Interests

The authors have no conflict of interests to disclose.

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