Case Report

A case report of extra-abdominal desmoid tumor after kidney transplantation

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Abbreviations & Acronyms
APC = adenomatous polyposis coli
COX-2 = cyclooxygenase-2
CT = computed tomography
DOX = doxorubicin
FAP = familial adenomatous polyposis
HE = hematoxylin eosin
MRI = magnetic resonance imaging
MTX = methotrexate
OPTN = organ procurement and transplantation network
SFT = solitary fibrous tumor
T2WI = T2-weighted image
VBL = vinblastine
WHO = World Health Organization

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Introduction: Desmoid tumors are rare, locally invasive, non-metastasizing intermediate grade neoplasms. According to the organ procurement and transplantation network report, approximately one million solid organ transplants have been performed globally, from which only three desmoid tumors have been reported.

Case presentation: We present the case of a 61-year-old male who developed an asymptomatic abdominal mass 3 years after kidney transplantation. Computed tomography revealed a 50-mm mass attached to the Hem-o-lok clip beneath the right rectus abdominis muscle. A needle biopsy revealed β-catenin positivity, which raised suspicion for a desmoid tumor. Due to progressive tumor enlargement, surgical excision was performed. The patient was discharged on postoperative day 5 without any surgical complications.

Conclusion: This is the first documented case of an extra-abdominal desmoid tumor after kidney transplantation. Although no recurrence has been observed over a 5-year follow-up period, careful long-term monitoring is required because of the high recurrence rate of desmoid tumors.

Key words: desmoid tumor, immunosuppressive therapy, kidney transplantation, surgery, β-catenin.

Keynote message

To our knowledge, this is the first report of an extra-abdominal desmoid tumor following kidney transplantation. The tumor presented an increase in size during observation, and surgical excision was selected as the treatment option, which was achieved with minimal damage to the adjacent organs and few surgical complications. The patient has remained recurrence-free 5 years after surgery, although continuous and careful follow-up is essential because of the high tumor recurrence rate.

Introduction

Desmoid tumors are classified as intermediate grade locally aggressive tumors that do not metastasize by the WHO guidelines. They have a rare rate of occurrence of 2.3–4.3 per one million people.[1] According to the OPTN report, approximately one million solid organ transplants have been performed globally; however, only three patients with post-transplant desmoid tumors have been reported.[2–4] For this reason, and there is limited information on the etiology, common sites of occurrence, cause of incidence, treatment options, recurrence rates, cause-specific prognosis, and the relationship between desmoid tumors and immunosuppressive therapy in transplant recipients.

Case presentation

A 61-year-old man with chronic renal failure and end-stage kidney disease due to autosomal dominant polycystic kidney disease received an ABO-compatible living donor kidney transplant from his wife. One month after surgery, the patient developed a pelvic lymphocele,

which was treated with laparoscopic fenestration using Hem-o-lok clip and remained recurrence-free during long-term follow-up. Three years after the transplantation, he noticed an asymptomatic abdominal mass. Physical examination revealed a nontender, firm, and immobile mass in the right lower abdomen, and plain abdominal CT revealed a 50 × 35 mm mass attached to the Hem-o-lok clip beneath the right rectus abdominis muscle (Fig. 1). MRI showed low intensity in the tumor on T2WI, and on fat-suppressed T2WI, the inside of the tumor showed a flow void (Fig. 2). CT and MRI showed possibly a SFT and desmoid tumor. Percutaneous needle biopsy of the mass using ultrasound sonography revealed the pathological finding of β-catenin positivity, thus suggesting a desmoid tumor. In the absence of FAP or related symptoms, the tumor was initially monitored using CT scans over a short period. The size of the tumor increased from 50 × 35 to 60 × 40 mm within 6 months, prompting surgical excision. During intraoperative findings, the tumor was found to firmly adhere to the Hem-o-lok clip, which had been used in the previous operation for post-transplant lymphocele. The resected specimen appeared as a yellowish-white irregular mass (Fig. 3), and the histopathological examination revealed a spindle cell proliferation, which stained positive for β-catenin and infiltrated the surrounding tissues (Fig. 4). Finally, an extra-abdominal desmoid was pathologically confirmed. The patient's postoperative course was uneventful, and he was discharged on postoperative day 5 without any surgical or medical complications.

Discussion

It has previously been reported that the etiology of desmoid tumors includes mutations in the *APC*, *FAP*, and β-catenin (CTNNB1) genes, post-surgical or traumatic responses, and the involvement of estrogen. The most common occurrence sites are the abdominal wall (49%), extra-abdominal sites

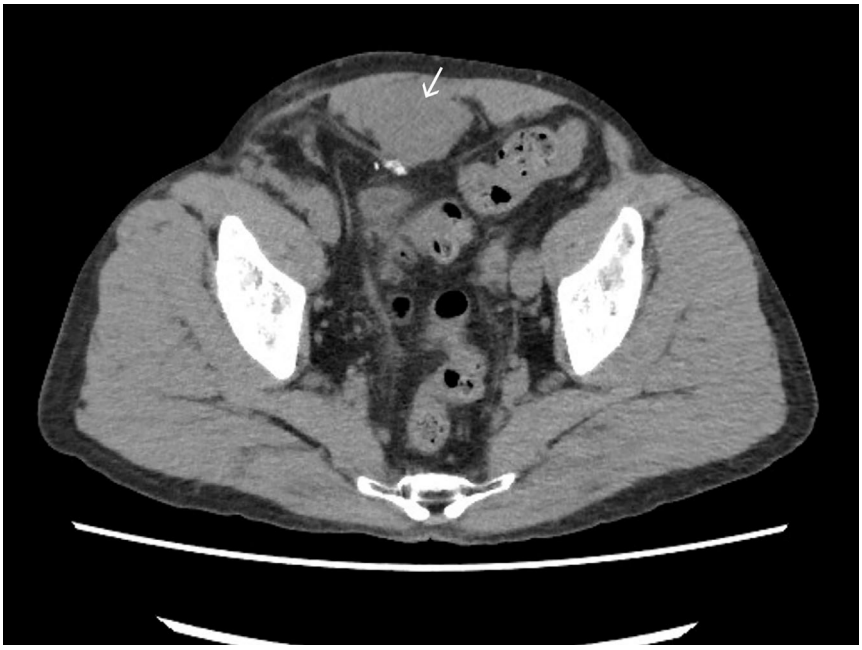

Fig. 1 Abdominal CT scan demonstrating a mass lesion of 50 mm × 35 mm under the right lower abdominal rectus muscle (arrow).

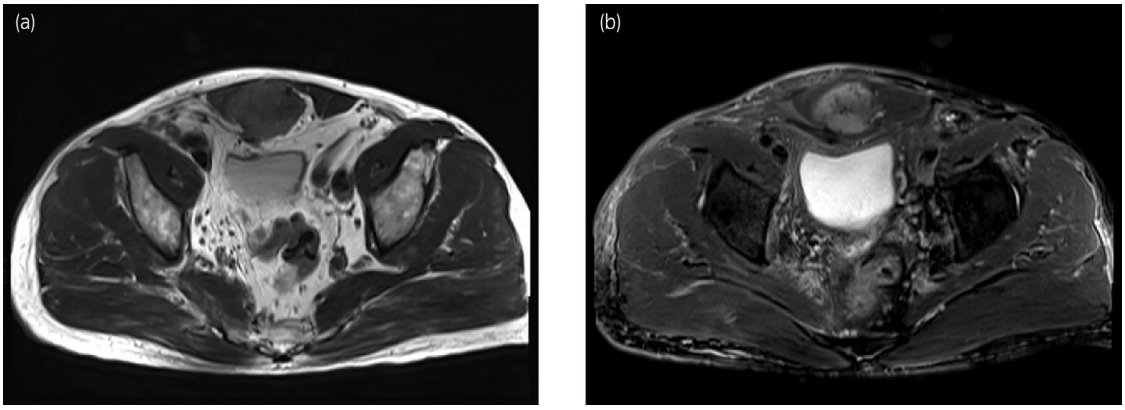

Fig. 2 (a) T2-weighted imaging. (b) Fat-suppressed T2-weighted imaging.

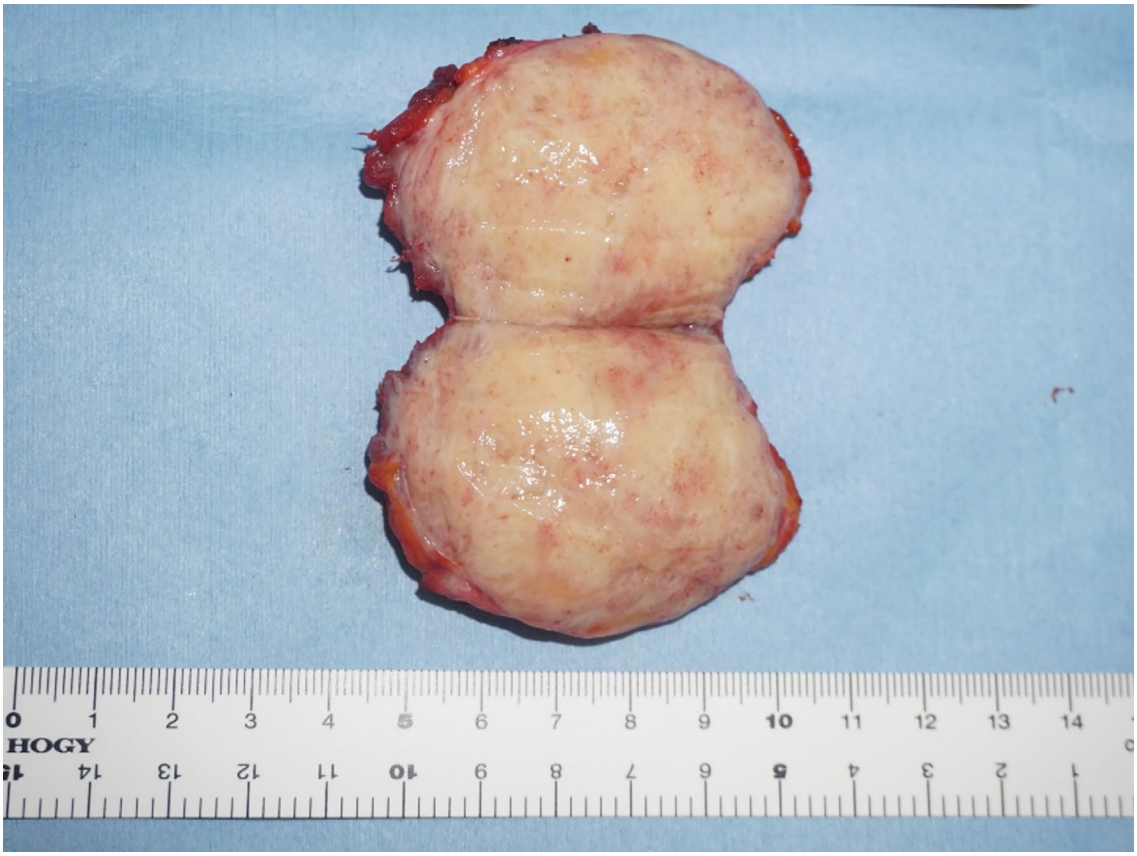

Fig. 3 The size of the surgical specimen was 50 × 40 × 30 mm, and the weight was 46 g.

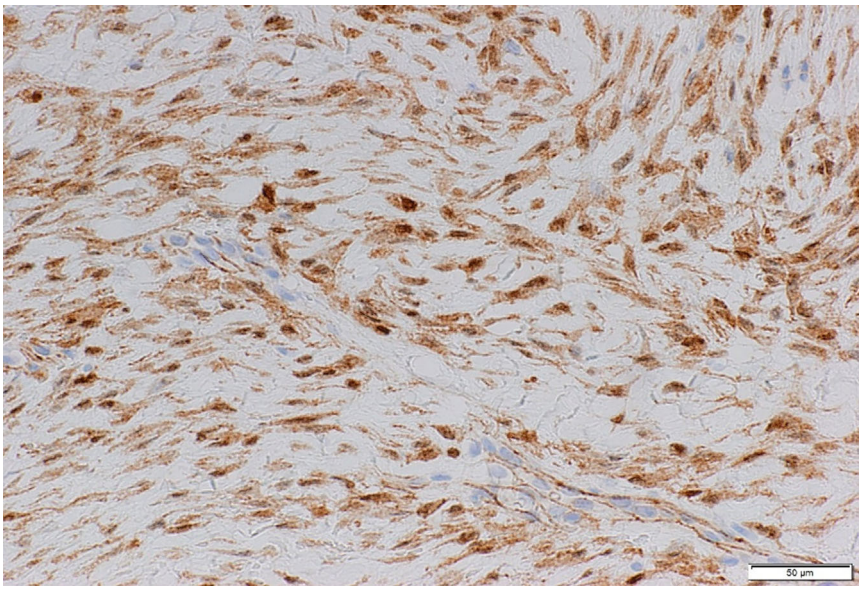

Fig. 4 The pathological findings were positive for β-catenin by immunohistochemical staining.

(43%), and intra-abdominal locations (mesentery, 8%).[1] Desmoid tumors occur in 2.3–4.3 people per million,[1] while approximately one million solid organ transplants have been performed according to the OPTN report. Among these, only three patients with desmoid tumors after solid organ transplantation have been reported, which indicates that this is the first documented patient with an extra-abdominal desmoid tumor after kidney transplantation. One previous patient was reported to have an intra-abdominal desmoid tumor that was unrelated to FAP following liver transplantation,[2] the second patient had an intra-abdominal desmoid tumor that was associated with FAP following kidney transplantation,[3] and the

third patient had a desmoid tumor that developed in the renal graft after kidney transplantation.[4] Similar to the current case, there are two reports of masses formed due to Hem-o-lok clip, both of which were operated on and diagnosed as granulomas.[5,6] Treatment strategies for desmoid tumors include observation, surgery, pharmacological therapies (COX-2 inhibitors, anti-estrogen therapy), radiation therapy, and chemotherapy (MTX + VBL, DOX).[7] Previously, Nakayama *et al*. reported spontaneous regression in three of 11 patients with desmoid tumors.[8] Among 194 patients with extra-abdominal desmoid tumors, 68% experienced recurrence following surgical excision, with recurrence rates of 48% after wide excision and 90% after marginal resection.[9] There have been no reports on the relationship between desmoid tumors and immunosuppressive therapy. In the present case, the patient had no family history of FAP but developed a lymphocele 1 month after kidney transplantation, which was treated with laparoscopic fenestration using Hem-o-lok clip for protection from recurrence of lymphocele, which might have been followed by the development of an extra-abdominal desmoid tumor. We initially selected an observation treatment option, based on suspected low recurrence risk. During the clinical course, an increase in tumor size with the absence of critical adjacent organs was detected on follow-up image analysis, and the patient then underwent surgical excision. Due to the high risk of recurrence caused by the nature of the tumor, close follow-up is required. Due to the rarity of desmoid tumors in transplant recipients, it remains unclear whether their incidence, treatment, and recurrence rates are consistent with those in non-transplant patients. Furthermore, the association between immunosuppressive therapy and desmoid tumor development is unknown, and further investigation with additional cases is needed to better understand the etiology of this rare disease.

Conclusion

We hypothesize that the development of the desmoid tumor in this patient may have been influenced by fenestration surgery using Hem-o-lok clip and the treatment procedure for a post-transplant lymphocele. To our knowledge, this is the first documented case of a patient with an extra-abdominal desmoid tumor after kidney transplantation. The tumor was excised because of its growth in the absence of adjunct critical organs and the reduced possibility of postoperative complications. The patient experienced no recurrence over the postoperative 5 years, and as desmoid tumors have high recurrence rates, careful follow-up is required after surgical excision.

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Author contributions

Tsuyoshi Matsuda: Writing – original draft. Shota Yamada: Writing – review and editing. Ayaka Tsuchiyama: Writing – review and editing. Hiroki Kurata: Writing – review and editing. Yuta Mukae: Writing – review and editing. Shinji Okano: Supervision. Yasushi Mochizuki: Writing – review and editing. Ryoichi Imamura: Supervision.

Conflict of interest

The authors declare no conflict of interest.

Approval of the research protocol by an institutional reviewer board

Not applicable.

Informed consent

We obtained informed consent from the patient.

Registry and the registration No. of the study/trial

Not applicable.

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