Case Report

Transcatheter arterial embolization of malignant pelvic solitary fibrous tumor: case report and literature review

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Abstract: Pelvic malignant solitary fibrous tumor (SFT) is a relatively rare disease, and literature on radical resection with transcatheter arterial embolization of pelvic SFT is lacking. In this work, we report on a 55-year-old man with a presacral mass who was hospitalized at our department. Computed tomography and magnetic resonance imaging indicated pelvic space-occupying lesions that were 12 cm × 10 cm in size and pelvic lesions that were not clearly demarcated from the right posterior wall of the bladder and the right ureter. This result suggested severe secondary hydronephrosis of the right renal pelvis. The patient underwent transcatheter iliac arterial embolization. Radical tumor resection was performed, and the results of pathological examination confirmed the diagnosis of malignant pelvic SFT. There was no SFT recurrence in this patient at 1-year follow-up. Herein, we report on the treatment of a patient with malignant pelvic SFT, a rare condition, who underwent successful radical resection after transcatheter arterial embolization. Transcatheter arterial embolization can block the blood supply of the SFT as much as possible and improve the possibility of tumor resection. In the future, pelvic SFTs can be considered improving the resection rate by transcatheter arterial embolization before surgery.

Keywords: Malignant solitary fibrous tumor; transcatheter arterial embolization; case report.

Submitted May 22, 2021. Accepted for publication Oct 20, 2021.
doi: 10.21037/tcr-21-887
View this article at: https://dx.doi.org/10.21037/tcr-21-887

Introduction

Solitary fibrous tumor (SFT) is a rare mesenchymal tumor mainly reported in the pleura (1). Recently, an increasing number of extrapleural SFTs, including pelvic SFTs, have been found in the clinical setting. Pelvic SFTs can be either benign or malignant. Malignant pelvic SFTs are extremely rare. The clinical diagnosis and treatment of malignant pelvic SFTs are sophisticated. Herein, we report on a case with malignant pelvic SFT and systematically reviewed the literature on pelvic SFTs. From our case, we put forward novel clinical experiment that transcatheter arterial embolization of malignant pelvic SFT could improve the tumor resection rate and prognosis. We present the following article in accordance with the CARE reporting checklist (available at https://dx.doi.org/10.21037/tcr-21-887).

Case presentation

A 55-year-old man presented to our hospital with persistent lower abdominal pain. The patient was otherwise healthy, with no family history of malignant tumors. Laboratory examination results were normal. Computed tomography

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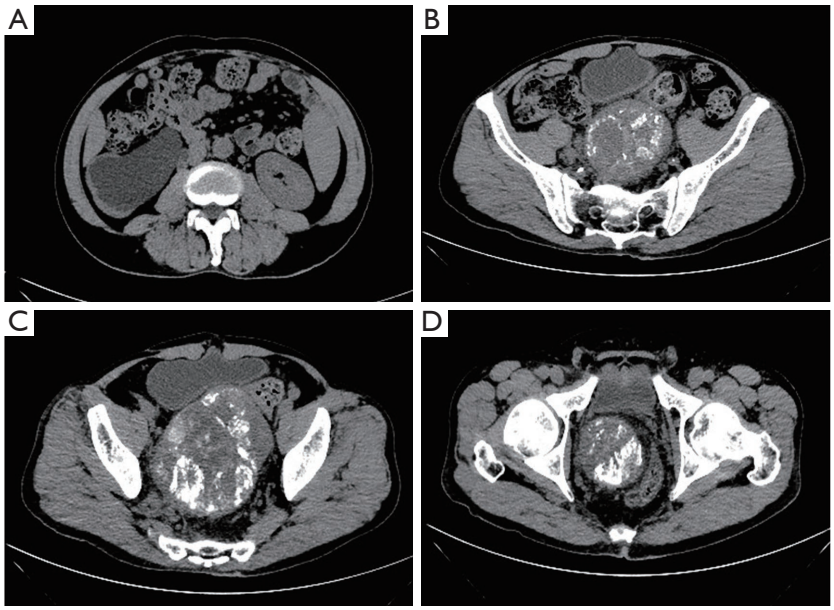

Figure 1 The CT examination results of different planes. (A) The sign of right hydronephrosis. (B-D) Different layers of the pelvic SFT. CT; computed tomography; SFT, solitary fibrous tumor.

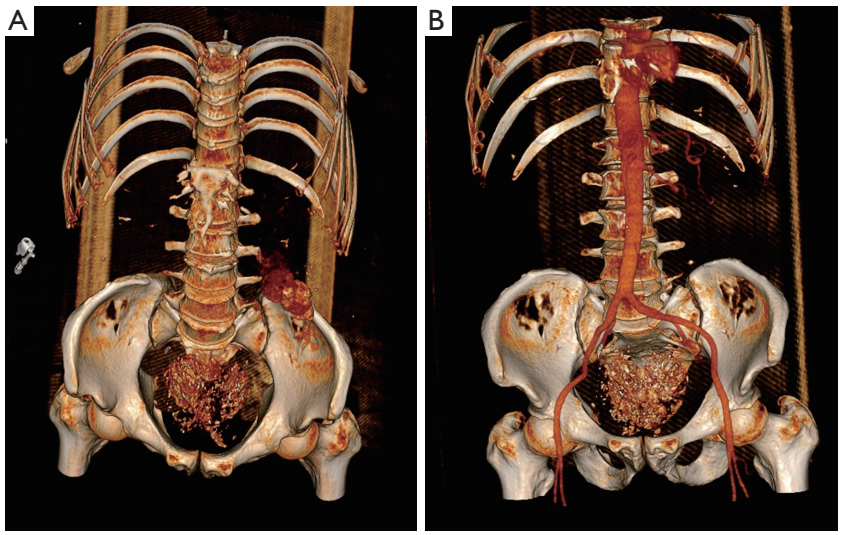

Figure 2 The three-dimensional reconstruction of the enhanced CT. (A) The CT three-dimensional reconstruction of pelvic SFT. (B) The CT three-dimensional reconstruction of pelvic SFT arteries. CT; computed tomography; SFT, solitary fibrous tumor.

(CT) and magnetic resonance imaging (MRI) indicated significant necrosis and calcification in the tumor.

Abdominal enhancement CT results indicated that there was a large mass of abnormal density behind the bladder. The pelvic mass was about 12.4 cm × 10.1 cm in size, and the CT value was 40 HU. There were multiple calcifications in the pelvic mass. Enhanced CT examination results showed slight enhanced density at the edge of the lesion (*Figure 1*). The 3D reconstruction of the enhanced CT is shown in *Figure 2*.

Pelvic MRI indicated significant abnormal signal shadows between the posterior bladder and rectum, about 12.4 cm × 9.1 cm × 8.1 cm in size (*Figure 3*). Inconsistent signals

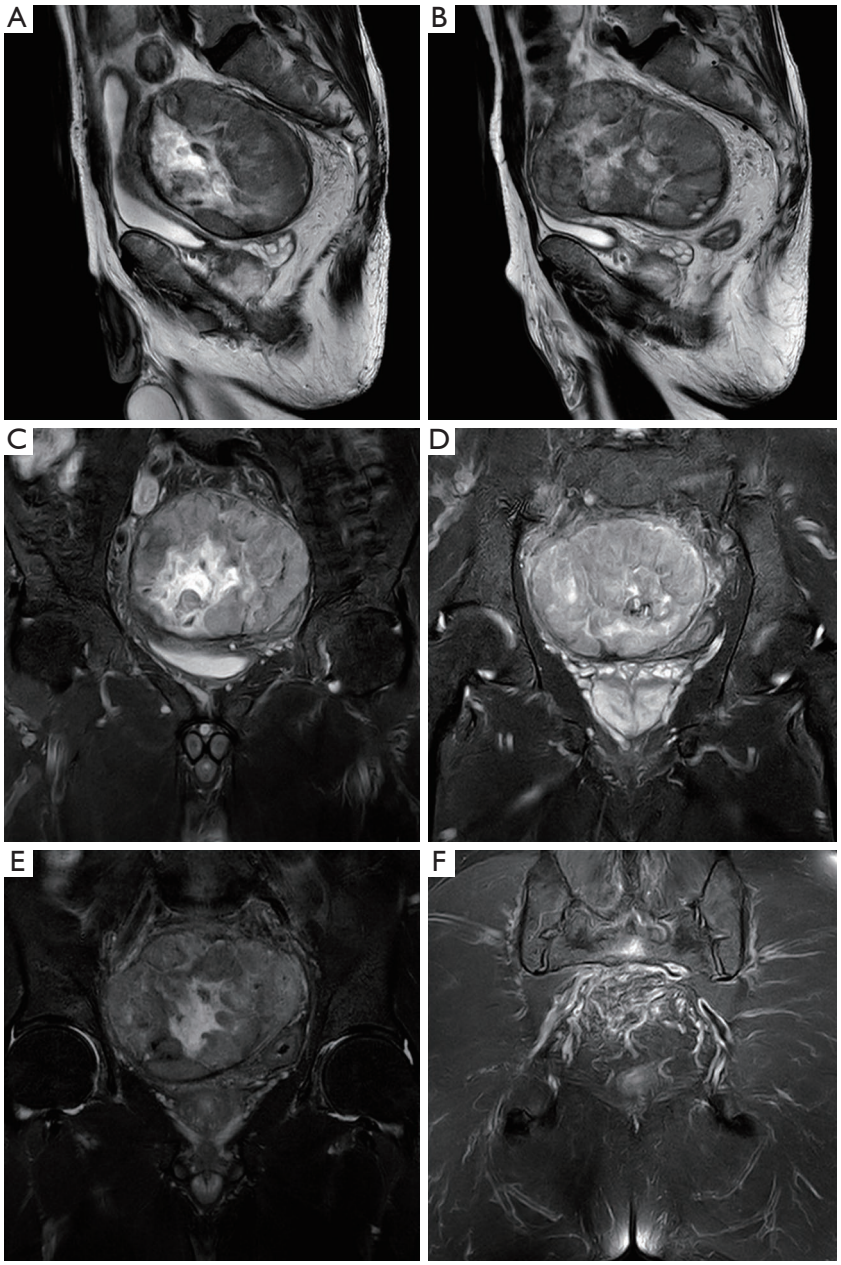

Figure 3 The MRI examination results of different planes and angles. (A,B) Sagittal plane MRI figure. (C-F) Different planes of pelvic posterior coronal MRI figure. MRI, magnetic resonance imaging.

were seen on long T1 and T2 images, and short T1 and T2 signals were seen in the lesion. A linear low-signal envelope could also be seen around the lesion. The lesion showed markedly uneven enhancement on enhanced examination. The edge of the lesion was also significantly enhanced. The boundary between the lesion and the right ureter was unclear, and the right ureter was obviously expanded. No obvious enlarged lymph node was found in the pelvic cavity.

The patient underwent transcatheter iliac arterial embolization, which obstructed the blood supply of the tumor. Tumor resection was performed, and the malignant pelvic SFT was completely removed within 1 hour.

The tumor envelope was intact, with some necrosis and calcification inside (*Figure 4A*). Hematoxylin and eosin

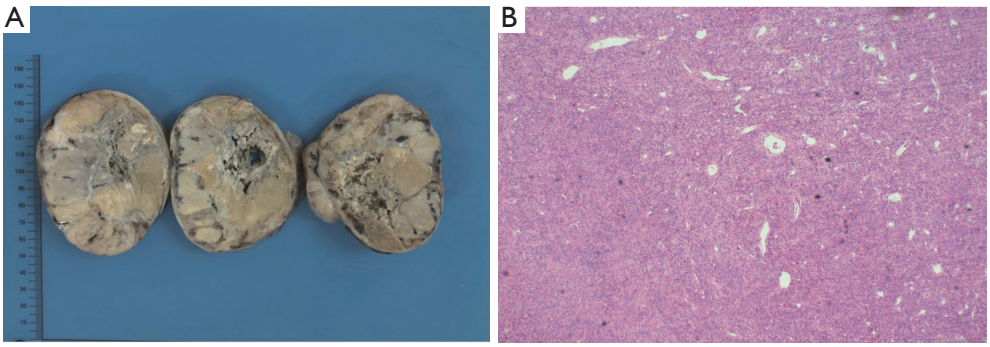

Figure 4 The general picture of SFT and H&E picture. (A) The photo of pelvic SFT of the patient. (B) The H&E result of pelvic SFT. Magnificent 200×. SFT, solitary fibrous tumor.

staining confirmed that that pelvic tumor was a malignant SFT with significant infarction (*Figure 4B*). As shown in *Figure 5*, the immunohistochemical results exhibited vimentin (–), cytokeratin (CK)-pan (focal positive), CD34 (+), CD99 (+), S-100 (–), B-cell lymphoma-2 (Bcl-2) (+), Ki-67 (+10%), P53 (+20%), CD31 (–), smooth muscle actin (–), h-Caldesmon (–), desmin (–), CD117 (–), Dog-1 (–), ALK (–), and STAT6 (+). Immunohistochemical results further confirmed the diagnosis of malignant pelvic SFT. The patient recovered well after the surgery and did not require adjuvant therapy. The patient did not accept postoperative radiotherapy or chemotherapy. The patient was followed up at 1 year, and had no recurrence of malignant SFT or lower abdominal pain symptoms. The patient was able to perform activities of daily living normally without discomfort.

All procedures performed in studies involving human participants were in accordance with the ethical standards of the ethics committee of the First Hospital of Jilin University and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Discussion

SFT, a rare mesenchymal tumor (2), was first reported in 1931 by Klemperer and Rabin (3). It was first described as a benign variant of mesothelioma (4). Although most SFTs are benign, about 20% of SFTs are malignant. The 2013 World Health Organization (WHO) classification of soft tumor defines SFTs as hypercellular, mitotically active tumors [>4 mitosis/10 high-power fields (HPF)], with cytological atypia, tumor necrosis, and/or infiltrative margins (5). The most effective treatment for SFT is complete resection. Furthermore, postoperative adjuvant chemotherapy for malignant SFT could prevent recurrence and metastasis.

The SFT can also exhibit various forms, including osteosarcomatous and chondrosarcomatous components (6), which makes the diagnosis of SFT challenging. Radiographic diagnosis is mainly dependent on CT and MRI. The CT values and MRI signal intensity of SFTs vary depending on the amount of collagen, vascular tissue, and myxoid and cystic degeneration in the tumor (7). Different types of SFTs exhibit varying manifestations on CT and MRI. However, Li *et al.* concluded that there was no difference in CT and MRI findings between malignant and benign SFTs (8); therefore, CT and MRI cannot be used to diagnose SFT. Fluorodeoxyglucose positron emission tomography (FDG-PET) can also be used as a supplementary examination for malignant or benign SFT. In previous cases, malignant SFTs showed increased FDG uptake, while the uptake of benign SFTs was negative (9). The clinical behavior of SFTs is hard to predict. Therefore, many SFTs are discovered after surgical treatment (10). Postoperative pathological diagnosis is the most reliable method for distinguishing SFT from other mesenchymal tumors and is the gold standard for the diagnosis for SFT. As a rare mesenchymal tumor, SFT exhibits as "patternless" ovoid and spindle cells, while it often has a unique immunohistochemistry staining results (positive for STAT6, CD34, CD99, and Bcl-2) (9). CD34, CD99, vimentin, and Bcl-2 constitute the basic pathological diagnosis of SFT (7), and most SFTs are positive for CD34, CD99, and Bcl-2 (2). The rearrangement of NGFI-A binding protein 2 (NAB2)–STAT6 can be found in almost all SFTs, which leads to

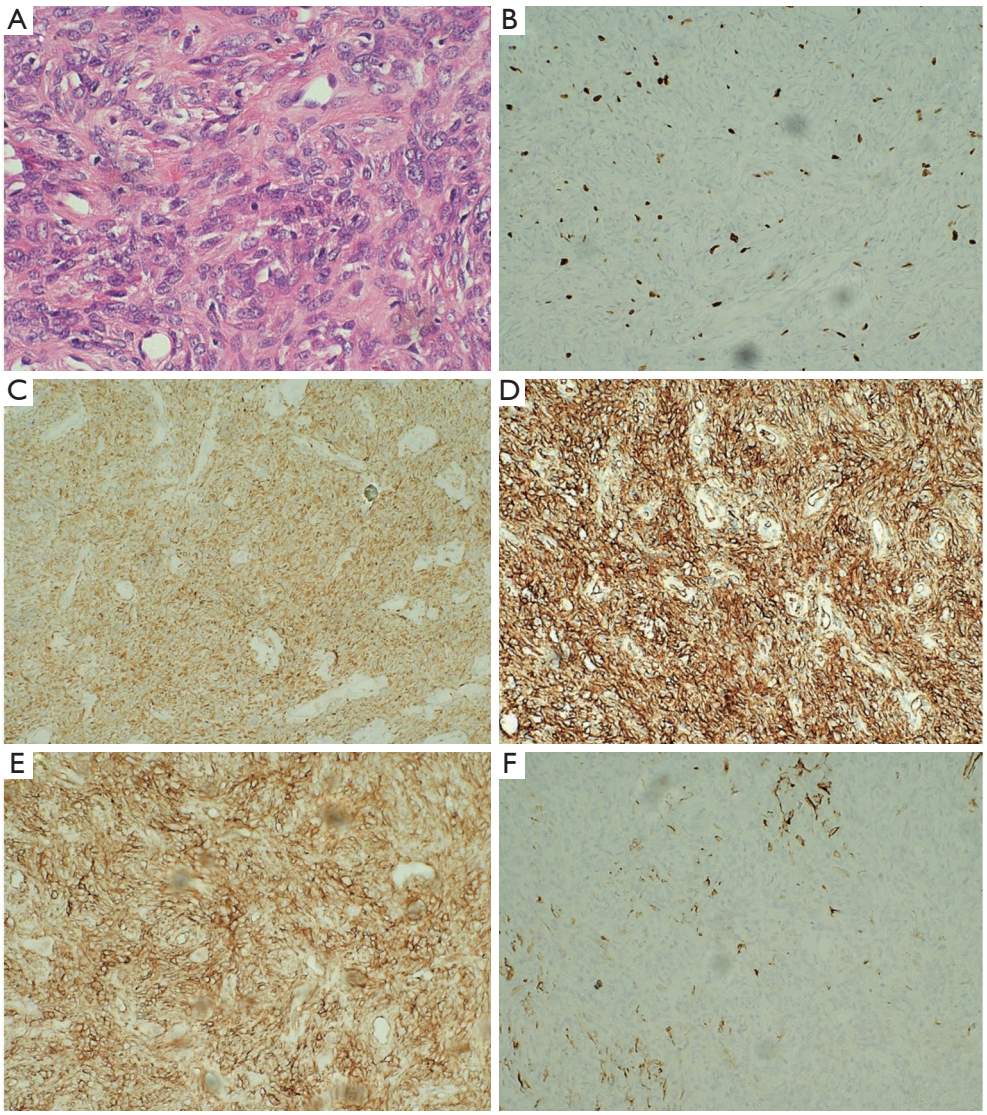

Figure 5 The immunohistochemical results. (A) H&E result of pelvic SFT; (B) Ki-67 result; (C) Bcl result; (D) CD34 result; (E) CD99; (F) CK-pan result. Magnificent 200×. SFT, solitary fibrous tumor.

the positive expression of STAT6 (11). STAT6 detection can increase the accuracy of SFT diagnosis. Currently, the rearrangement of NAB–STAT6 is considered as specific histological diagnostic standard for SFT. SFTs can undergo dedifferentiation transformation, and this particular type of SFT is difficult to distinguish from Ewing's sarcoma, therefore making SFT diagnosis difficult (12). Awareness of immunohistochemical and molecular biological characteristics can help to accurately diagnose SFT.

Malignant SFTs are rare, and there are no established criteria to diagnosis malignant SFTs (13). In their study, England *et al.* concluded the criteria for malignant SFTs (14). As shown in *Table 1*, malignant SFTs have the following characteristics: (I) hypercellularity (>4 mitoses/10 HPF), (II) nuclear pleomorphism, (III) necrosis, (IV) tumor infiltrative growth, and (V) tumor size >10 cm. Although there are obvious differences between benign and malignant SFTs, some SFTs are incorrectly characterized as benign. Retrospective studies have shown that patients with benign pathological SFTs may also have recurrence after resection (5). This can be attributed to the limitations of pathological diagnosis. For indistinguishable SFTs, total

Table 1 The systematic review of pelvic SFT case reports

Reference	Gender	Location	Age	Pathology	Treatment	Author
(15)	Female	Pelvic; Broad ligament	37	Malignant	Tumor resection, sub-extensive hysterectomy, bilateral salpingo-oophorectomy, and pelvic lymphadenectomy with chemotherapy	Chen *et al.*
(16)	Female	Uterine cervix/ left parametrium	45	Benign	Radical abdominal hysterectomy with bilateral salpingo-oophorectomy and pelvic lymphadenectomy	Nowakowski *et al.*
(7)	Male	Seminal vesicle	68	N	Laparoscopic seminal vesicle tumor resection	Zhao *et al.*
(17)	Male	Bladder serosa	41	Malignant	Partial cystectomy	Dozier *et al.*
(18)	Male	Urinary bladder	67	Malignant	Partial cystectomy and segmental resection	Cheng *et al.*
(19)	Female	Urinary bladder	24	Benign	Partial cystectomy with total resection of the remaining tumour tissue	Heinzelbecker *et al.*
(20)	Female	Urinary bladder	59	Benign	Radical cystectomy	Tzelepi *et al.*
(21)	Male	Urinary bladder	60	Benign	Radical prostatectomy with complete tumor excision	Leite *et al.*
(2)	Male	Urinary bladder	49	Malignant	Complete surgical resection	Prunty *et al.*
(6)	Female	Intra-Pelvic SFT	70	Malignant	Complete surgical resection	Kurisaki-Arakawa *et al.*
(3)	Female	Presacral SFT	52	Benign with focally malignant feature	Laparoscopic surgical resection	Kim *et al.*
(22)	Female	Pelvic mesorectum SFT	27	Malignant	Trans-sacral tumor resection	Soda *et al.*
(23)	Male	Prostate	35	Benign	Radical prostatectomy with partial excision of the bladder wall	Oguro *et al.*
(24)	Male	Prostate	21	Benign	Epicystotomy	Grasso *et al.*
(25)	Male	Seminal vesicle	56	Benign	Complete surgical resection	Funahashi *et al.*
(4)	Female	Uterine cervix	68	Benign	Robotic-assisted radical hysterectomy, bilateral salpingo-oophorectomy, and pelvic lymph node dissection	Rahimi *et al.*
(26)	Male	SFT of Pelvic	52	Benign	Embolization followed by surgical resection	Boe *et al.*
(27)	Male	Pelvic peritoneum	61	Malignant	Laparotomy tumor resection	Vossough *et al.*
(10)	Male	Prostate	46	N	Trans Urethral Resection Prostate then nerve-sparing retropubic radical prostatectomy	Yang *et al.*
(28)	Female	Pelvic SFT	53	Benign	Surgical resection	Zhao *et al.*
(29)	Male	Pelvic SFT	52	Malignant	Radical excision	Yan *et al.*
(30)	Male	Pelvic SFT	71	Malignant	Radical prostatectomy and partial rectal excision	Ando *et al.*
(31)	Male	Pelvic SFT	64	N	Surgical resection	Tsushimi *et al.*
(13)	Male	Prostatic urethra	68	Malignant	Surgical resection	Tanaka *et al.*
(32)	Male	Pelvic pre-rectal tumor	69	N	Previous embolization and en bloc resection	Garcia-Amador *et al.*

Table 1 (*continued*)

Table 1 (*continued*)

Reference	Gender	Location	Age	Pathology	Treatment	Author
(33)	Female	Mesorectum	56	Malignant	Laparoscopic tumor resection	Kawamura *et al.*
(34)	Female	Perianal tumor	56	Benign	Trans-sacral tumor resection	Katsuno *et al.*
(35)	Male	Pelvic tumor	52	Malignant	Pelvic tumor resection	Gao *et al.*

N means not mentioned. SFT, solitary fibrous tumor.

Table 2 Criteria for malignant pelvic SFT

Malignant SFT characteristic
1. Presence of hypercellularity (>4 mitoses/10 HPF)
2. Nuclear pleomorphism
3. Necrosis
4. Tumor infiltrative growth
5. Tumor size >10 cm

SFT, solitary fibrous tumor; HPF, high-power fields.

resection is necessary. Therefore, detailed preoperative preparation and complete resection of tumors are the best treatments for SFT.

SFT mainly occurs in the pleura. In their single center study, DeVito *et al.* found that the primary SFT site was the lung/pleura in 28 (34%) patients, the abdomen/pelvis in 23 (28%), the extremities in 13 (16%), and the head/neck in 9 (11%). Pathology was benign in 42 (51%) patients and malignant in 40 (49%) (5). SFTs originating in the pelvis have been reported to be more asymptomatic and more aggressive than SFTs originating in the pleura (12), and about 5–10% patients with SFT outside the pleura exhibit hypoglycemia (15,36). As shown in *Table 2*, pelvic SFTs can be classified as bladder SFT, prostate SFT, seminal vesicle SFT, uterine cervix SFT, pelvic SFT, or peritoneum SFT. *Table 1* summarizes our systematic review of pelvic SFTs. SFTs can also develop in the bladder, seminal vesicle, cervix, and prostate. For pelvic SFT, tumor resection is the first choice of treatment.

Pelvic SFTs are mostly hypervascular, and thus a lack of preparation for radical resection may cause massive hemorrhaging (3). Therefore, it is important to reduce bleeding during operation. In their study, Soda *et al.* indicated that pelvic SFTs should be carefully prepared prior to resection (22). Kayani *et al.* performed successful surgical resection after embolization of the pelvic SFT (12), while Garcia-Amador *et al.* found that embolization of the median sacral artery and branches of the internal iliac arteries is necessary to facilitate surgery (32). Pelvic tumors, especially pelvic SFTs, are usually supplied by the internal iliac arteries (34). Due to an abundant blood supply and the deep location of tumors, malignant pelvic neoplasms are often difficult to operate on. Transcatheter embolization is an effective treatment for pelvic tumors. In their study, Pisco *et al.* confirmed the efficacy of preoperative transcatheter embolization for pelvic tumors (37). This was also confirmed in our case: preoperative artery embolization markedly reduced intraoperative hemorrhage, alleviated tumor adhesion, and shortened the operative time. Embolization of internal iliac arteries obviously reduced the operative time and promoted recovery after operation.

There is no standard curative treatment for pelvic SFT other than surgery. Intra-arterial chemotherapy and glucocorticoid therapy are reported to be effective for malignant insulin-like growth factor (IGF-2) secreting SFTs. Furthermore, tyrosine kinase inhibitors, such as imatinib and sunitinib, have also been reported to be effective therapies for unresectable pelvic SFTs (38). It is important to prevent recurrence of pelvic SFT after resection. Therefore, guidelines for the adjuvant therapy of pelvic malignant SFTs are urgently needed in the future.

In conclusion, malignant pelvic SFT is a rare mesenchymal carcinoma. Here, we reported on a patient with a malignant pelvic SFT. The patient indicated the first symptom to be abdominal pain. The patient underwent internal iliac artery embolization in preparation for surgery, and the tumor was completely resected. Finally, the patient recovered well with no recurrence 1 year after transcatheter arterial embolization and surgery.

The take-away concept provided in this case is that transcatheter arterial embolization could be effectively block blood supply of pelvic SFT. After transcatheter arterial embolization, the pelvic SFT is easier to be removed and the got better prognosis.

Acknowledgments

Funding: This work was funded by the Youth Foundation of The First Hospital of Jilin University (No. JDYY102019001) and the Science and Technology Development Program of Jilin Province (No. 2020122256JC).

Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at https://dx.doi.org/10.21037/tcr-21-887

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://dx.doi.org/10.21037/tcr-21-887). LH reports that this work was supported by Youth Foundation of The First Hospital of Jilin University (No. JDYY102019001) and The Science and Technology Development Program of Jilin Province (No. 2020122256JC). The other authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in studies involving human participants were in accordance with the ethical standards of the ethics committee of the First Hospital of Jilin University and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

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Cite this article as: Yan X, Zheng C, Wang J, Li D, Lu J, He L, Wang C. Transcatheter arterial embolization of malignant pelvic solitary fibrous tumor: case report and literature review. Transl Cancer Res 2021;10(11):4979-4987. doi: 10.21037/tcr-21-887

