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Case Report
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A Case Report of Malignant Pelvic Solitary Fibrous Tumor and Literature Review

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Abstract

A 47-year-old man visited the genitourinary outpatient department complaining of difficulty with urination and defecation for several weeks. A pelvic tumor was found incidentally in abdominal sonography. Further study using abdominal computed tomography and Magnetic Resonance Imaging (MRI) demonstrated a large heterogenous mass with central necrosis over the pelvic cavity that was compressing the bladder and prostate anteriorly. Exploratory laparostomy with removal of the pelvic tumor was performed without complications. Pathology examination revealed a solitary malignant fibrous tumor with dedifferentiation at intermediate risk. The patient received adjuvant radiation therapy postoperatively without any evident complication or recurrence at 6-month follow-up.

Keywords: Malignant pelvic solitary fibrous tumor; Surgical intervention; Adjuvant therapy

Introduction

Solitary fibrous tumor is a spindle cell neoplasm mostly originating from the pleura; however, Occurrence at other anatomic sites, such as the mediastinum, retroperitoneum, pelvis, and meninges, has also been reported. Although this tumor is usually considered an indolent neoplasm, the prognosis is unpredictable and related to morphological features [1]. Here, we present a case with Malignant Pelvic Solitary Fibrous Tumor (MPSFT) and review the literature.

Case Presentation

Patient information

A 47-year-old male had difficulty with urination and defecation for several weeks and sought treatment. One pelvic tumor was found incidentally *via* abdominal sonography (Figure 1). Further study using Computed Tomography (CT) and Magnetic Resonance Imaging (MRI) revealed one lobulated mass, measuring 9.0 cm × 6.4 cm × 8.6 cm and exhibiting central necrosis, located in the pelvic cavity and in close contact with the posterior wall of the urinary bladder and prostate (Figures 2, 3).

Surgical and pathological findings

Exploratory laparostomy *via* low abdominal incision with removal of the pelvic tumor was performed uneventfully (Figure 3). No severe adhesion and easy division of the tumor mass from the adjacent organs and tissues (bladder, prostate, and seminal vesicle) was noted during surgery.

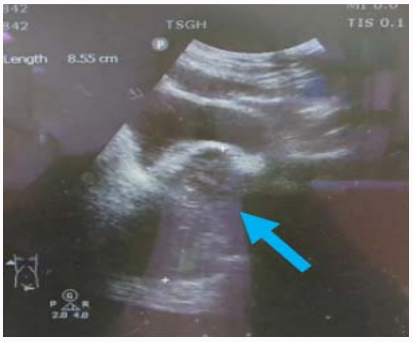

Figure 1: Pelvic sonography indicated a hypoechoic lesion compressed upon bladder posterior wall (arrow).

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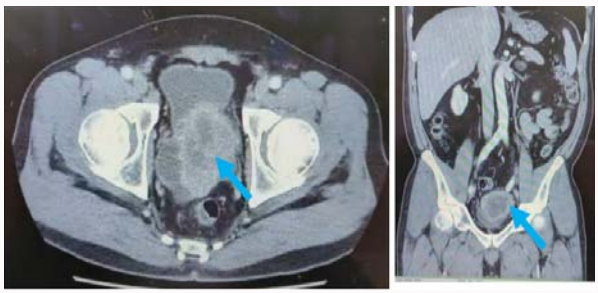

Figure 2: CT scan indicated a heterogenous mass located in the pelvis with central necrosis and contact with the bladder and left seminal vesicle.

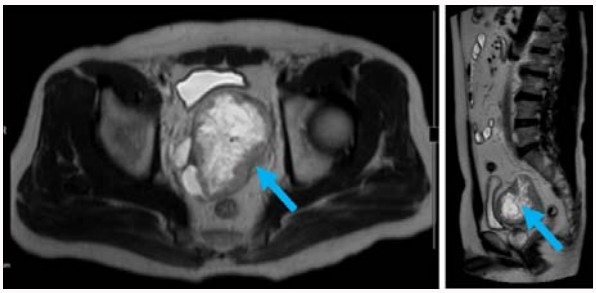

Figure 3: MRI demonstrated a large lobulated mass in the pelvic cavity, which appeared to be of hyperintense density in the central area on T2WI (arrow).

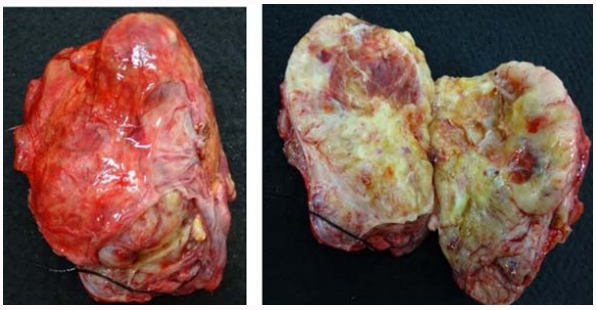

Figure 4: Gross pictures of the resected pelvic tumor; its cutting surface revealed pale whitish tumor tissue with central necrosis in content.

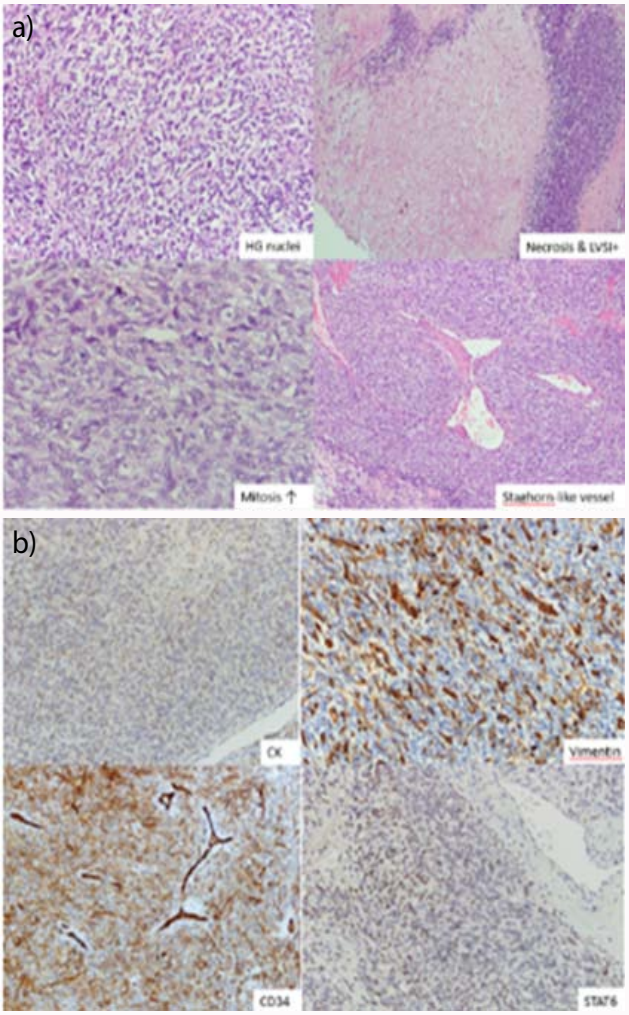

Figure 5: (a) High grade nuclei, necrosis, Lymphovascular Space Invasion (LVSI) positive, high level mitosis, and staghorn-like vessels were noted in the resected tumor tissue (H&E staining, 100x). **(b)** Positive immunohistochemical staining for vimentin, CD34, and STAT6; negative for CK in the resected tumor tissue (400x).

The tumor was covered by a pseudocapsule, and the pathology examination with specific immunohistochemical stains revealed the tumor to be a MPSFT with dedifferentiation and posing an intermediate risk (stage: pT2N0M0) (Figures 4, 5).

Follow-up and outcomes

The patient was referred to the radiotherapy department for postoperative adjuvant radiation therapy with a full dose of 45 Gy in 25 fractions. As of 6-month follow-up, no tumor recurrence has occurred.

Discussion

A previous study reported that only 6% of all SFTs originate from the pelvis. SFTs are usually benign, but 12% to 22% of them are malignant. In recent 30 years, a total of 26 patients with MPSFT without secondary transformation to malignancy have been reported presenting with lower urinary tract or gastrointestinal symptoms (Table 1). Most patients received surgical resection, and one of them underwent biopsy examination to confirm the diagnosis. Such tumors typically occur in patients aged 40 to 70-years, without a difference in gender distribution. The follow-up period ranges from 0.5 to 8 years. Fourteen of 26 patients documented in the literature experienced a tumor relapse, three patients succumbed to cancer, and the remaining patients were alive or lost to follow-up [1-18].

With regard to diagnosis, CT images of MPSFTs usually demonstrate a highly vascular tumor lesion with persistence of enhancement in the arterial, portal venous, and delayed phases, and approximately 20% to 30% of tumors develop calcification. In MRI, the tumor typically presents a low signal on T1W1 and high signal on T2W1. Frequently, these image patterns appear similar to a gastrointestinal stromal tumor or leiomyosarcoma. Although the real nature of a pelvic tumor cannot be accurately defined by images, resection or biopsy maybe considered as the first choice.

Histologic diagnostic features of MPSFT should be considered if one or more of the following criteria exist: 1) high cellularity, 2) high mitotic activity (more than 4 mitoses per 10 high-power fields), 3) pleomorphism, 4) necrosis, and 5) hemorrhagic changes [19]. In our case, the patient's histological feature showed a typical fibrous tumor with high mitotic activity and necrosis. Therefore, an intermediate risk of MPSFT was inferred.

Table 1: Summary of patients with MPSFT in the literature.

Author	Gender	Age	Max Size (cm)	Management	Tumor Relapse	Management Post Relapse	FU Time	Last Status FU
He et al.	M	46	3.8	Surgery	Y (1 y)	Surgery + RT	-	NA
Mitra et al.	M	55	10.3	Surgery	N	-	4 m	AWD
Lu et al.	M	41	14	Surgery	Y(5 m)	Sunitinib	8 m	AWD
Ronchi et al.	M	62	20	Surgery	N	-	8 y	AWD
Ishihara et al.	M	72	7	Surgery	Y(12 y)	Surgery	-	NA
Dozier et al.	M	41	28	Surgery	N	-	8 m	AWD
Baldi et al.	M	52		Surgery	Y(10 y)	Surgery	2 y	DOD
Baldi et al.	M	31	-	Surgery	Y(10 y)	Surgery	0 y	AWD
Baldi et al.	M	61	-	Surgery	Y(11 y)	Surgery	2 y	DOD
Baldi et al.	F	66	-	Surgery	Y(11 y)	Surgery	1 y	LFU
Shoji et al.	M	41	12.9	Surgery	N	-	1 y	AWD
Kawamura et al.	F	74	9	chemoembolization therapy	Y(3 m)	RT	1 y	AWD
Nagase et al.	M	60	14	Surgery	N	-	-	AWD
Vossough et al.	M	61	9	Surgery	N	-	-	AWD
Mora-Guzmán et al.	F	83	7.5	Surgery	N	-	3 y	AWD
Kim et al.	F	52	12	Surgery	N	-	3 y	AWD
Kurisaki-Arakawa et al.	F	70	17	Surgery	N	-	4 m	DOD
Li et al.	M	59	10.2	Surgery	Y(-)	Surgery	-	NA
Li et al.	M	61	12	Surgery	Y(-)	Surgery	-	NA
Li et al.	M	47	11.3	Surgery	Y(-)	Surgery	-	NA
Li et al.	M	57	4.7	Surgery	N	-	-	NA
Thanasak et al.	F	54	16	Surgery	Y (3 y, 13 m, 12 m)	Surgery+ CCRT+ bevacizumab	5 y	AWD
Wagner et al.	M	28	18	bevacizumab + Surgery	N	-	-	AWD
Anne-Valerie et al.	F	32	-	Surgery + RT	Y (6 y)	-	7 y	AWD
Anne-Valerie et al.	M	71	20	Surgery + Chemotherapy	Y (1 y)	-	2 y	AWD
This case	M	47	9	Surgery	N	-	6 m	AWD

Effective adjuvant therapy for MPSFT after surgical treatment has yet to be established. Postoperative radiotherapy has been performed in some cases, with no recurrence or disease progression reported at follow-up [9]. However, no standard chemotherapeutic regimens for MPSFT are currently available. Combination therapy with temozolomide and bevacizumab was reported to be effective in some patients with MSFT [20]. Adjuvant target therapy (tyrosine kinase inhibitors), such as pazopanib, sorafenib, and sunitinib, is under clinical trial for MPSFT treatment [21-23].

In conclusion, MPSFT is a rare clinical entity. Surgeons must be alert to it as a possible diagnosis to enable them to initiate early surgical resection in patients presenting with pelvic tumor.

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