Volume 19, No 2 | International Journal of Radiation Research, April 2021

Solitary fibrous tumor of the urinary bladder in an 85-year-old woman: Case report and review of literature

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► ***Case report***

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Revised: January 2020
Accepted: April 2020

Int. J. Radiat. Res., April 2021; 19(2): 465-471

DOI: 10.29252/ijrr.19.2.465

ABSTRACT

A solitary fibrous tumor of the urinary bladder is an extremely rare type of mesenchymal tumor. We present a case of an 85-year-old woman with a large (14.5×10.7×10.1 cm) protruding intravesical mass detected by computed tomography (CT) and magnetic resonance imaging (MRI), which was proven to be a solitary fibrous tumor through histopathological examination. The clinicopathologic and radiological features are here discussed, along with a review of the published literature on this topic. Solitary fibrous tumors of the urinary bladder are more common in men (76%), with a peak incidence in people 40–60 years old. Most such tumors show heterogeneous intensity with a complete capsule. A decrease in the T2WI signal with an increase in collagen tissue content and gradual enhancement on CT/MRI images are their main features. Most such tumors are positive for CD34 under immunohistochemistry (100% in 24 available cases). The majority of tumors (88%) studied have been non-malignant and most patients (95.5%) showed favorable prognosis after a long-term follow-up.

Keywords: *Solitary fibrous tumor, urinary bladder, computed tomography, magnetic resonance imaging, histopathological.*

INTRODUCTION

A solitary fibrous tumor (SFT) is a rare spindle cell tumor of the mesenchymal origin. It accounts for under 2% of all soft-tissue tumors that usually involve the pleura. An extra-pleural SFT of the urinary bladder is an extremely rare type of mesenchymal tumor [1, 2]. We here present a new case of an SFT arising in the urinary bladder, in the oldest patient yet reported case, and we review 24 cases published in pertinent works from 1997 on, focusing on the importance of preoperative diagnosis (table 1) [1-21].

Case history

An 85-year-old woman presented with dysuria and hesitancy lasting two months. During physical examination, a movable firm mass over the lower abdomen was noted. Subsequent pelvic computed tomography (CT) and magnetic resonance imaging (MRI) showed a protruding intravesical mass with a complete capsule arising from the trigone of the urinary bladder. CT revealed a well-defined, oval-like solid mass measuring 14.5 × 10.7 × 10.1 cm (figure 1a). MRI showed the mass to be isointense in T1-weighted images (T1WI) (figure 1b), of heterogeneous intensity with a thin low-signal capsule in T2-weighted images (T2WI) (Figure 1c), and slightly hyperintense under diffusion-weighted imaging (DWI). The tumor showed map-like, gradual enhancement on MRI contrast-enhanced images (figure 1d-f).

Partial cystectomy was used for complete resection of the tumor. An elliptical gray-white tumor that showed a smooth surface upon gross inspection was resected, measuring 14×12×6 cm (figure 2a). There was a congestion on the surface of the mass, which was dark red, about

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[DOI: 10.52547/ijrr.19.2.27]

Table 1. Reported 25 cases of SFT in the urinary bladder: clinical and surgical features.

Case No.	Reference	Age	Gender	Symptoms	Size (cm)	Immunohistochemistry (positive)	Non-malignant / malignant	Treatment	Follow-up (months)
1	Bainbridge 1997[3]	50	Female	Urinary frequency	5.2	CD34, CD99, SMA, MSA, desmin	Non-malignant	TURBT	18
2	Bainbridge 1997[3]	42	Male	Pelvic pressure	20	CD34, CD99	Non-malignant	Wide excision	6
3	Westra 2000 [4]	67	Male	Asymptomatic, incidental cystoscopy finding	4	CD34	Malignant	Cystoprostatectomy	9
4	Westra 2000 [4]	67	Male	Asymptomatic, incidental MRI finding	NA	CD34	Non-malignant	TURBT	1
5	Corti 2001[5]	50	Male	Pelvic pain, dysuria, urinary bleeding	6.5	CD34, bcl-2, vimentin, IGF-Ⅱ mRNA	Non-malignant	Cystectomy	24
6	Kim 2004[6]	56	Male	Voiding difficulty, frequency, residual urine sensation	12	CD34, bcl-2	Non-malignant	Wide excision	12
7	Leite 2004[7]	60	Male	Incidental MRI finding for prostate adenocarcinoma	3.2	CD34, bcl-2	Non-malignant	Wide excision	11
8	Saint-Blancard 2006[8]	38	Male	Dysuria, palpable abdominal mass	15	CD34, vimentin, bcl-2	Non-malignant	Cystectomy	30
9	Tzelepi 2007 [9]	59	Female	Intermittent hematuria	8.5	CD34, vimentin, CD99, CD68	Non-malignant	Cystectomy	77
10	Heinzelbecker 2008[2]	24	Female	Hematuria	8.5	CD34, bcl-2, vimentin, CD99	Non-malignant	Partial cystectomy	24
11	López Martín 2010[10]	59	Male	Voiding difficulty, frequency, residual urine sensation	4	CD34, bcl-2, vimentin, CD99	Non-malignant	Wide excision	24
12	Wang 2010 [11]	50	Male	Hematuria	8	CD34, vimentin	Non-malignant	Wide excision	9
13	Bruzzone 2010[12]	74	Male	Chills, diaphoresis, acute abdominal pain, hematuria	10	NA	Non-malignant	Wide excision	NA
14	Seike 2012 [13]	41	Female	Incidental finding	5.2	CD34, bcl-2	Non-malignant	TURBT	NA
15	Cheng 2012 [14]	67	Male	Persistent pain over lower abdomen	16	CD34, CD99	Malignant	Partial cystectomy	18
16	Wang 2012 [15]	72	Male	Incidental MRI finding	8.5	CD34, bcl-2	Non-malignant	TURBT	16
17	Otta 2014 [16]	78	Male	Hematuria	NA	CD34, bcl-2, vimentin, CD99	Non-malignant	TURBT	5
18	Spairani 2014[17]	60	Male	Voiding difficulty	9	CD34, bcl-2, vimentin	Non-malignant	Partial cystectomy	11
19	Dozier 2015 [18]	41	Male	Weight loss, progressive abdominal bloating	28	CD34, bcl-2, vimentin	Malignant	Partial cystectomy	8
20	Mustafa 2016[1]	36	Female	Urgency, occasional straining to void, increased abdominal girth	10	CD34, bcl-2, vimentin, CD99	Non-malignant	Cystectomy	NA
21	Tanaka 2016 [19]	60	Male	Incidental Ultrasound finding	8	CD34, Ki-67, STAT6	Non-malignant	Wide excision	24
22	Tanaka 2016 [19]	60	Male	Incidental Ultrasound finding	4	CD34, CD99, c-kit, Bcl-2	Non-malignant	Partial cystectomy	120
23	Lhungay 2017[20]	70	Male	Worsening urinary incontinence, urgency	9	CD34, bcl-2, vimentin, CD99	Non-malignant	TURBT	12
24	Urbina-Lima 2019[21]	61	Male	Hypoinsulinemic hypoglycemia	23	CD34, vimentin	Non-malignant	Partial cystectomy	12
25	Present case	85	Female	Dysuria, hesitancy	14	CD34, CD56	Non-malignant	Partial cystectomy	6

TUBRT, transurethral resection of the bladder; TURP, transurethral resection of prostate; NA, not available.

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[DOI: 10.52547/ijrr.19.2.27]

3×4 cm in size, and the rest of the tumor was smooth (figure 2b). Microscopically, cellular areas with spindle-shaped or ovoid cells arranged in pattern-less among sparse collagen fibers were observed. These cells had poorly defined cytoplasm, homogeneous nuclei and inconspicuous nucleoli (figure 3a-c). Immunohistochemical analysis showed the tumor cells to have positive expression of CD34 (figure 3d) and slight positive expression of CD56 (figure 3e), but no expression of CD31 (figure 3f), HMB45, Chromogranin A, Cytokeratin (CK), CK20, CK7, SMA, Synaptophysin and S-100. Based on histology and immunohistochemistry, the neoplasm established the diagnosis of SFT. There was no evidence of tumor recurrence during a 6-month follow-up.

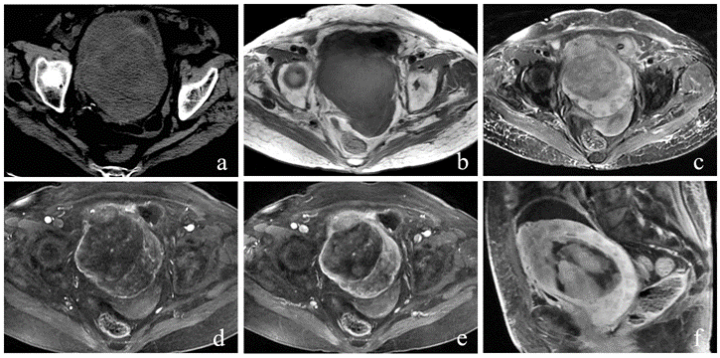

Figure 1. (a) CT revealed a large protruding intravesical mass with a complete capsule and components of cystic. **(b-f)** MRI revealed an elliptical mass in the urinary bladder which was isointense on T1WI, heterogeneous intensity on T2WI, and accompanied by gradual enhancement.

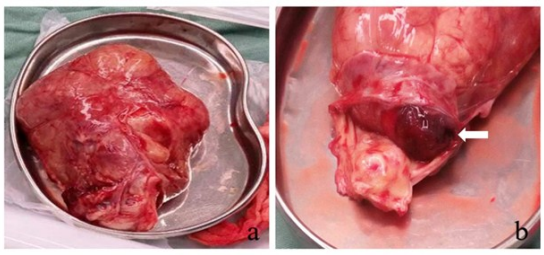

Figure 2. (a) Gross picture of the resection specimen showed a well-circumscribed mass measuring 14×12×6cm. **(b)** There was a congestion on the surface of the mass, which was dark red, about 3×4 cm in size (White arrow).

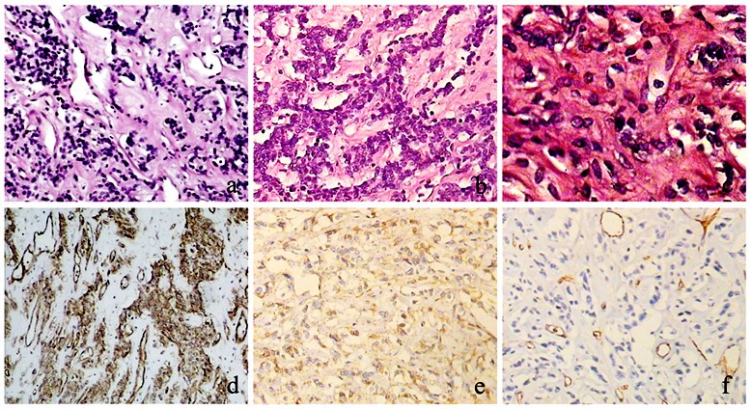

Figure 3. (a-c) Hematoxylin and eosin stained section showed a spindle cell proliferation in a dense collagenous stroma. (H&E, 100×, 200× and 400×, respectively). **(d-f)** Immunohistochemistry of CD34, CD56 and CD31, respectively (DAB, 200×). Staining pattern confirmed SFT.

DISCUSSION

The SFT was first recognized as a distinct neoplasm by Klemperer and Rabin in 1931, which was initially described in the pleura [22]. SFTs usually involve the pleura, pericardium, and peritoneum, but they are extremely rare in the urinary bladder, with the first case reported in 1997 [3]. To our knowledge, only 24 cases of SFT in the urinary bladder have been reported to date. We reviewed the 24 cases and summarized their clinicopathological and imaging features in tables 1 and 2 to emphasize the importance of preoperative diagnosis.

According to published data, including those of our patient, SFT of the urinary bladder is more common in men (76%), in whom the age of onset ranged from 24 to 85 years (mean age,

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Table 2. Imaging features of the reported cases of SFT arising in the urinary bladder.

Case No	Reference	Age	Gender	Imaging features	Location
1	Corti 2001 [5]	50	Male	Echography, CT and X-ray urography showed a well-circumscribed pelvic mass.	Between prostate and urinary bladder
2	Kim 2004 [6]	56	Male	Ultrasonography revealed a heterogeneous echogenic intravesical mass. CT scans showed about a 12cm sized protruding intravesical mass without perivesical infiltration.	The dome of the bladder
3	Leite 2004 [7]	60	Male	MRI identified a tumoral mass measuring 3.0cm, homogeneous, well delimited, hyperintense in T2WI, showing intense enhancement following injection of contrast agent.	Close to the left lateral bladder wall
4	Saint-Blancard 2006[8]	38	Male	Ultrasound revealed a heterogeneous bladder mass about 15 cm in diameter. Intravenous urography found a laterovesical tumor without hydronephrosis.	The trigone of the urinary bladder
5	Heinzelbecker 2008[2]	24	Female	At excretory urography, the upper urinary tract on the left side was slightly dilated, and the indwelling catheter was displaced to the left. CT revealed a large tumor filling the urinary bladder with suspected connection to the tumor of the right ovary.	The left side wall of the bladder
6	Wang 2010 [11]	50	Male	CT showed a protruding intravesical mass without perivesical infiltration, 7.5 cm × 7.1 cm in size, and the internal density was not uniform.	The right lateral wall of the bladder
7	Cheng 2012 [14]	67	Male	MRI of the pelvis revealed a large lobulated soft-tissue solid tumor. The hyperintense cystic components were considered to indicate areas of central necrosis.	The dome of the urinary bladder
8	Spairani 2014[17]	60	Male	CT scans confirmed a protruding intravesical mass without perivesical infiltration, 7.6 × 6 cm in size.	Occupying a large section of vesical volume
9	Dozier 2015 [18]	41	Male	CT scan revealed a 26.8 × 21cm intra-abdominal mass occupying most of his abdominal cavity.	Abutting the superior dome of bladder
10	Mustafa 2016[1]	36	Female	MRI showing large heterogeneous solid T2 signal intensity nodule in the pelvis measuring 6.7 × 7.5 × 7.2 cm with iso-enhancement of the uterine parenchyma and appears to be connection to left aspect of cervix.	The left lateral bladder wall
11	Tanaka 2016[19]	60	Male	CT revealed a well-delineated and homogeneous retrovesical solid mass that measured 5 × 3 cm in diameter. MRI confirmed this noninfiltrative solid tumor, with a thin capsule of low signal in T2WI.	The posterior wall of the bladder
12	Lhungay 2017[20]	70	Male	CT scan with and without contrast showed a large, 8.0 × 6.7 × 7.2cm, heterogeneous enhancing mass that encompassed most of the left half of the bladder and appeared to be invading through the bladder wall, with possible prostatic invasion.	The left lateral wall and trigone area of the bladder
13	Urbina-Lima 2019 [21]	61	Male	CT: voluminous abdominal-pelvic mass, predominantly solid, of 15 × 23 × 22 cm compressing adjacent structures	The posterior bladder wall
14	Present case	85	Female	CT revealed a well-defined solid mass with mixed density. It was isointense on T1WI, heterogeneous signal on T2WI, and a slightly hyperintense on DWI, accompanied by heterogeneous and gradual enhancement during CT/MRI contrast enhancement.	The trigone of the urinary bladder

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57.08 years), and the peak incidence was 40–60 years old. SFTs of the urinary bladder were usually asymptomatic at first. Urinary tract irritation, hematuria, pelvic compression, and other symptoms can develop as the tumor grows. Some tumors secrete insulin-like growth factor II, which can lead to hypoglycemia [5, 21, 23]. In our review of 24 cases, including our case, 8 (32%) of the patients had difficulty voiding, 7 (28%) incidental imaging findings, 7 (28%) discomfort of the lower abdomen, and 6 (24%) presented with hematuria.

The SFT of the urinary bladder often presented as a well-defined elliptical soft tissue mass, and a large tumor volume with an average size of 10.4 cm (range from 3.2 to 28 cm). It usually appeared as heterogeneous density on CT. For this reason, cystic and hemorrhagic areas were visible inside the tumor. The map-like heterogeneous enhancement and gradual enhancement during CT contrast enhancement were helpful to the preoperative assessment of tumoral vascularity and indicated regions of tumoral hemorrhage and cystic degeneration. Intra-tumoral calcification was rare but not specific. MRI showed isolated lesions with clear boundaries. These were isointense or slightly hypointense on T1WI and isointense or of heterogeneous intensity on T2WI. Cystic and mucosal components showed a high signal on T2WI. T2WI signal decreased as collagen tissue content increased, a characteristic finding of SFT. Gradual enhancement and the delayed enhancement were the most significant, which was its main feature [24, 25]. According to our review, 13 of 24 cases with available imaging data showing that all tumors had clear boundaries and heterogeneous intensity, which contributed to the diagnostic process (table 2). In our case, the neoplasm showed heterogeneous intensity on T2WI. This and the hypointensity on T2WI especially suggested collagen tissue content, and the tumor showed distinct gradual enhancement, all of which were consistent with the typical findings of SFTs.

However, the final diagnosis of an SFT involving the urinary bladder relies on histopathological features. Grossly, the SFT is a round solid mass with a smooth boundary. It is grayish-white to tan due to the vascular and collagen content. Hemorrhage and cystic degeneration can occur in the tumor. Microscopically, the tumor is composed of spindle cells (hypercellular areas) and hyaline collagen (hypocellular areas), which are usually arranged haphazardly. The alternate distribution of the cell's sparse and rich regions was characteristic of an SFT. Immunohistochemistry of CD34 (90%–95%), CD99 (70%), and bcl-2 was mostly positive, and CD34 is recognized as a relatively specific and accurate immunological marker [5], which is positive in all of the 24 available reported cases. Recent studies have demonstrated that immunohistochemical analysis of NAB2-STAT6 is positive in up to 100% of SFTs and can be used for confirmatory diagnostic test of SFT [26, 27]. Unfortunately, NAB2-STAT6 analysis was not performed in our case.

The treatment of choice is complete resection with negative margins. It has a favorable prognosis with long-term survival rate, and five-year overall survival approaches 100% [28]. Most SFTs of the urinary bladder (80%–90%) are non-malignant with indolent clinical courses, but can potentially recur or metastasize. Positive surgical margins, tumor diameter greater than 10 cm, and poor histology were associated with aggressive behavior. Some studies established the criteria for malignancy in SFTs including tumor size (≥10 cm), mitotic count (≥ 4/10 HPFs), high tumor cellularity, high nuclear pleomorphism, and tumor necrosis. The mitotic count (≥ 4/10 HPFs) was the only independent prognostic factor. Scores based on these factors stratify patients to low, medium or high risk and have significant prognostic value for recurrence during follow-up [29]. However, only 3 cases (12%) of malignant SFT have been reported with an average diameter of 16 cm (4 cm, 16 cm, 28 cm, respectively) [4, 14, 18]. Simultaneously, 23 of the 25 cases had tumor size information available, and the average size was approximately 10.4 cm (range: 3.2 to 28 cm). Recently, some scholars have used Imatinib for the treatment of SFTs, to good effect [30]. Similar studies have brought hope for targeted therapy

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[DOI: 10.52547/ijrr.19.2.27]

of SFTs. Follow-up information was available in 22 patients (88%), with an average follow-up time of approximately 22 months (range: 1 month to 120 months). However, 3 cases presented with aggressive behavior but no tumor recurrence. Only 1 case of tumor recurrence occurred in among the remaining cases of non-malignant tumors, reported by Otta *et al.* [16]. Although most SFTs have favorable prognoses, long-term follow-up is necessary to determine the behavior of an SFT in the urinary bladder.

CONCLUSION

An SFT of the urinary bladder is an extremely rare type of mesenchymal tumor. We here describe a female patient with an SFT originating in the urinary bladder. This is the oldest patient reported. Some characteristic features of CT and MRI contribute to preoperative diagnosis. The treatment of choice is complete resection with negative margins. Long-term follow-up is necessary for this patient to ensure no local recurrence, which we recommend for all cases of SFTs.

Statement of Ethics

The materials in the manuscript were obtained strictly according to the ethical standards. Informed consent of the patient was obtained.

ACKNOWLEDGMENTS

This study has received funding by grants from the National Natural Science Foundation (81471652, 81771824, 81701681).

***Conflicts of interest*:** *Declared none.*

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