



Solitary fibrous tumor involving urinary bladder: a case report and literature review

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Abstract: Solitary fibrous tumor (SFT) is a rare fibroblast stroma tumor involving the mediastinum and pleura. We herein describe an SFT of bladder which is extremely rare and review 29 similar cases in the last decades. We present a case of a 52-year-old male patient who suffered from urinary urgency and frequency for 12 months. Non-contrast computed tomography (CT) showed a slightly high density calcified mass with 70 mm × 61 mm in diameter. Contrast-enhanced CT demonstrated the mass was slightly enhanced. Cystoscopy revealed a huge mass with flat surface. Histopathological review of the biopsy specimens could not confirm the diagnosis. Partial cystectomy was then performed and the diagnosis of SFT was confirmed by immunohistochemistry. The patient is doing well at 12 months follow-up without recurrence and metastasis. In conclusion, the diagnosis of SFT involving bladder should combine clinical presentation and imaging features. Complete surgical resection is the primary method and long-term follow-up is necessary.

Keywords: Case report; review; solitary fibrous tumor (SFT); urinary bladder.

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Introduction

Solitary fibrous tumor (SFT) is a rare fibroblast stroma tumor involving the mediastinum and pleura (1). The tumor is the most common diagnosed between 40 to 70 years old and the incidence rate is the same in males and females. SFT most commonly occur in the pleura and abdomen, only few cases of SFTs of bladder have been reported in the literature. The aim of this article is to present this rare case and review 29 similar cases reported in the literature in the last decade. We present the following case in accordance with the CARE Guideline (2).

Case presentation

A 52-year-old male Asian patient had suffered from urinary

urgency and frequency since August 2017. Therefore, he visited the local hospital and received simple urine analysis, which showed lower urinary tract infection. Then he was recommended to take levofloxacin to control the infection. The symptoms were partially relieved, but persisted and went worse in a year. He then accepted abdomen non-contrast CT, which revealed a slightly high-density calcified mass with 70 mm × 61 mm in diameter suspected as bladder cancer, locating on the right wall of bladder. He was transferred to our department. After admission, further contrast-enhanced CT was done and it demonstrated the mass was slightly enhanced and invading the bladder wall (*Figure 1*). Next, the patient underwent cystoscopy and it revealed a huge mass with flat surface, but the panorama could not be accurately manifested due to serious intravesical bleeding and the small bladder capacity

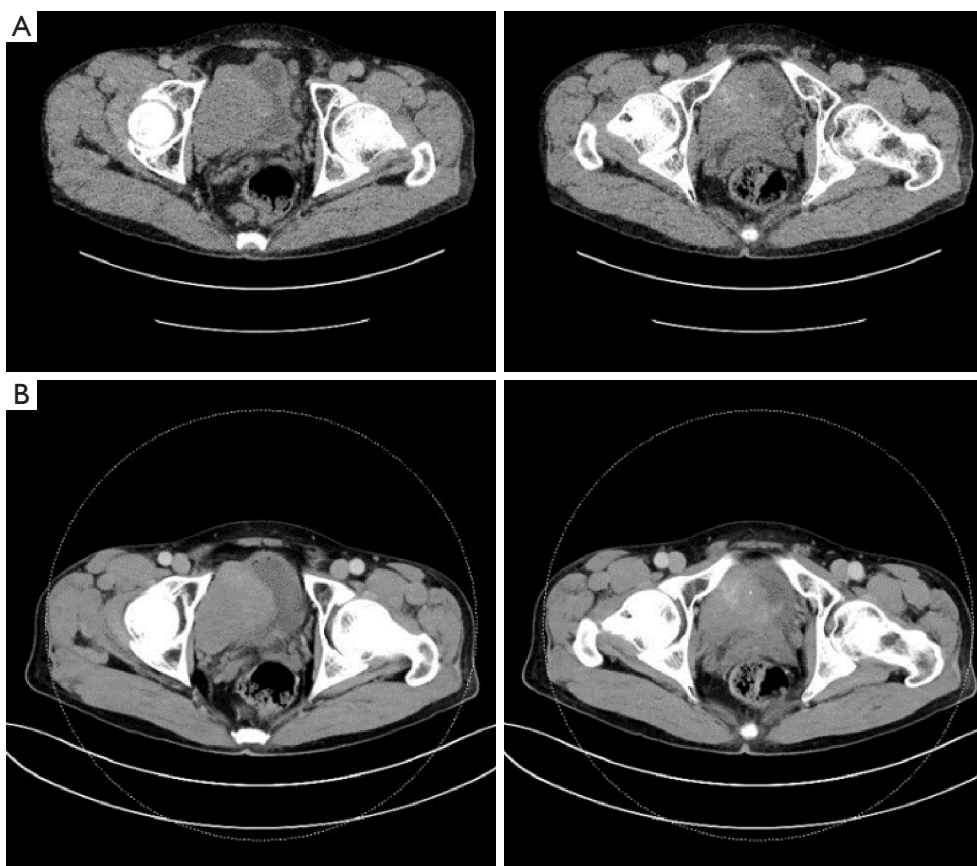


Figure 1 Preoperative CT. (A) Non-contrast CT showed a slightly high-density mass, calcification inside. (B) Enhanced CT demonstrated the mass was slightly enhanced.

that patient was hard to bear during the examination. Histopathological review of the biopsy specimens could not confirm the diagnosis. Finally, partial cystectomy was recommended. he had an unremarkable medical, surgical, and family history. On admission, his physical examination and laboratory data were also unremarkable. Preoperative CT evaluation did not find evidence of metastasis in both lungs and abdominal organs.

In the surgery, we chose a lower abdominal median incision to expose the bladder step by step. With a longitudinal incision of the bladder, we could see the bulge of the bladder mucosa on the right which is about 5 cm in diameter. Then we removed the tumor completely including 1cm of normal bladder tissue around, during which the bilateral ureteral orifices were recognized and protected by the ureteral catheters. We finally closed the bladder retaining bladder stoma, pelvic drainage tube and

urethral catheter which were removed 5, 6 and 7 days separately after the surgery. The patient tolerated the procedure well and there were no adverse events. The IHC (immunohistochemistry) showed expression of CD34, Bcl-2, PR, Ki67 (+2%), and negative for CK-pan, SMA, Desmin, DOG-1, ALK p80, S-100, thus the tumor was diagnostic as SFT (*Figure 2*). After 12 months of follow-up, the symptoms of the patient were significantly relieved. The abdominal CT (*Figure 3*) confirmed no signs of recurrence and distant metastasis (*Figure 4*).

Discussion and review of the literature

Epidemiology and origin

SFT was first described in 1931 by Klemperer (3). Originally, SFT was described as the tumor involving the mediastinum and pleura. However, with the increasing

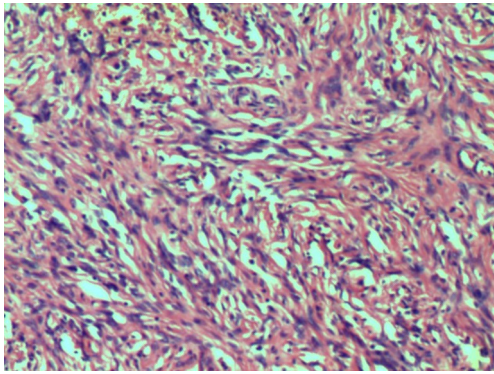


Figure 2 The tumor was mainly composed of spindle cell. Based on hematoxylin and eosin staining. Magnification: ×20.

reports in recent years, it has been established that SFT can occur widely in many organs including peritoneum, retroperitoneal, pericardium, gastrointestinal tract, kidney, adrenal gland, paranasal sinuses, orbit and so on. SFT arises from dendritic interstitial cells with positivity for CD34. It is a kind of non-directed differentiated stromal tumor, which can differentiate into fibroblasts, myofibroblasts, vascular cortical cells and vascular endothelial cells (4). According to the literature reports of 160 patients with SFT in the abdomen, until 2010, 28 were found in the liver, 25 in the prostate, 20 in the kidney, 12 in the retroperitoneal, and only 10 in the bladder (5). In view of its rarity, we are reporting this case to date and review of 29 cases of SFT

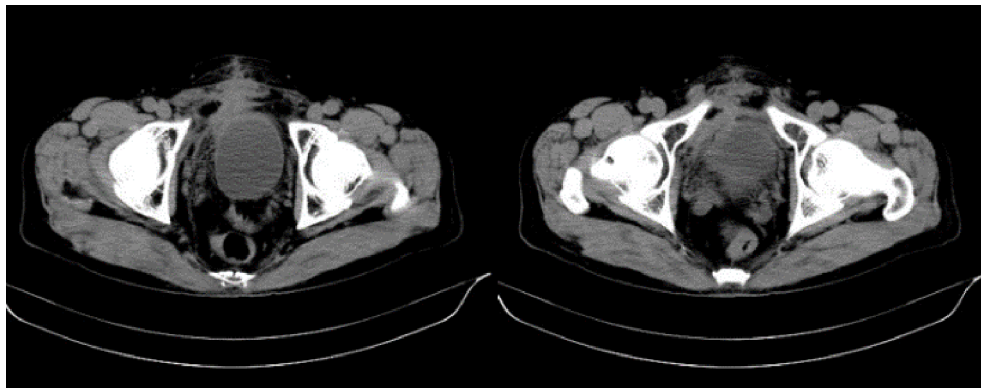


Figure 3 Abdomen non-contrast CT showed that the bladder capacity had partially recovered 12 months after operation.

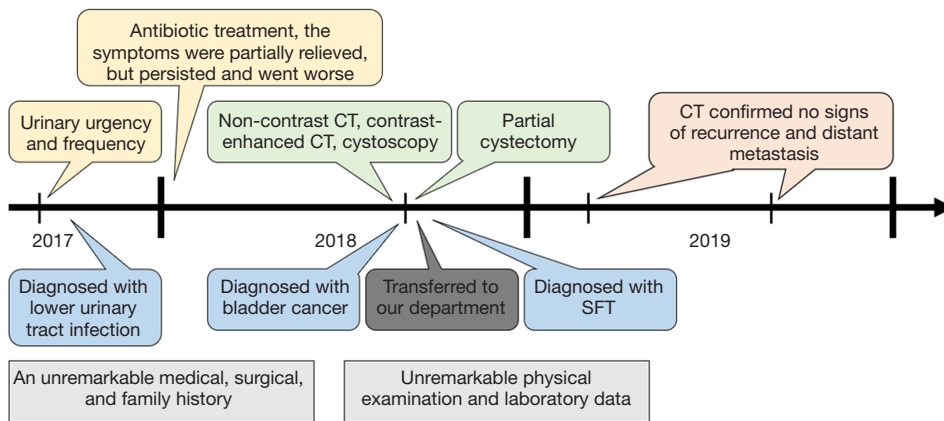


Figure 4 The timeline of the disease course.

involving bladder (Tables 1,2).

In our review, the age of onset of SFT patients was 11 to 85 years, while the mean age was 56.8 years and the median age was 60 years. The sex ratio with the patients is 7 females to 23 males, revealing a male predilection.

Diagnosis

Clinical feature was mostly related to tumor volume. Symptoms may occur when tumor volume is large and compresses surrounding tissues or organs. The most common symptoms were abdominal pain (9/28, 32.1%) and dysuria even urine retention (9/28, 32.1%), followed by hematuria (7/28,25%), urinary urgency (6/28, 21.4%), urination pain (5/28, 17.9%), abdominal swelling or palpable mass (5/28,17.9%), gastrointestinal symptoms (3/28, 10.7%), hypoglycemia (3/28,10.7%), weight loss (2/28, 7.1%) and 5 patients (5/28,17.9%) were incidental finding. Tumor sizes have ranged from 0.85 to 28 cm, while

the mean sizes were 10 cm.

On imaging, ultrasonic diagnostics generally presents as hypoechoic mass, clear boundary and uneven internal echoes, which is non-specific. Color Doppler showed blood flow signals in some cases. It is worth mentioning that the SFT favors malignancy, if hemodynamics shows low resistant index (RI) (31). CT or magnetic resonance imaging (MRI) helps to identify SFT and decide operation method. Yet, the features of CT or MRI are non-specific. Larger SFT was isodense with some hypodense areas caused by necrotic, myxoid or cystic changes on unenhanced CT while small SFT was isointense in both on CT and MRI (32). Some studies consider that rich blood vessels are reliable signs of SFT (33).

Due to the absence of specific clinical manifestations special characters in iconography and exclusive tumor markers, this disease is neglected frequently in the early stage. Currently, histopathology and immunohistochemistry are the main evidence for the diagnosis of SFT. With the

Table 1 Case reports of literature review (patient information)

Case No.	Reported year	Authors (reference)	Age (years)	Gender	Presentation	Size (cm/cm ² /cm ³)	Contour of tumor	Treatment
1	2010	Bruzzone <i>et al.</i> (5)	74	Male	Chills, diaphoresis, acute abdominal pain, hematuria	10×8×7	Smooth	Surgical resection
2	2010	Pata <i>et al.</i> (6)	76	Male	A low abdominal pain, acute urine retention, constipation	17×10×9	N/A	Surgical resection
3	2010	Wang <i>et al.</i> (7)	50	Male	Terminal gross hematuria, residual urine sensation, urination pain	3×5×8	Smooth	Surgical resection
4	2012	Cheng <i>et al.</i> (8)	67	Male	A palpable mass in right lower abdomen, pain during exercise	16×9×9	Smooth	Partial cystectomy and segmental resection of the intestine
5	2012	Seike <i>et al.</i> (9)	41	Female	Incidental US finding, residual urine sensation	5.2×3.4	Smooth	Transurethral resection
6	2012	Wang <i>et al.</i> (10)	72	Male	Incidental MRI finding	0.85	N/A	Transurethral resection
7	2014	Otta <i>et al.</i> (11)	78	Male	Hematuria, acute urinary retention	N/A	N/A	Transurethral resection
8	2014	Spairani <i>et al.</i> (12)	60	Male	Urination and pelvic pain	9×7×5	Smooth	Surgical resection
9	2016	Tanaka <i>et al.</i> (13)	60	Male	Incidental US finding	8×6	Smooth	Surgical resection
10	2016	Tanaka <i>et al.</i> (13)	60	Male	Incidental US finding	4×4	N/a	Surgical resection
11	2014	Tian <i>et al.</i> (14)	66	Male	N/A	7.9	Smooth	N/A

Table 1 (Continued)

Table 1 (Continued)

Case No.	Reported year	Authors (reference)	Age (years)	Gender	Presentation	Size (cm/cm ² /cm ³)	Contour of tumor	Treatment
12	2014	Chen <i>et al.</i> (15)	40	Female	Incidental physical examination finding	5×5	Smooth	Laparoscopic partial resection of bladder tumor
13	2015	Aji <i>et al.</i> (16)	60	Female	Progressive worsening of the flow of urine	1.2×0.9	N/A	Transurethral resection
14	2015	Dozier <i>et al.</i> (17)	41	Male	Abdominal pain, abdominal fullness, constipation, urinary frequency, a 25-pound weight loss	28×21×18	Smooth	Surgical resection
15	2015	Gao <i>et al.</i> (18)	52	Male	A mass in the right lower quadrant	9.3×6.5	Not smooth	Pelvic tumor resection; postoperative radiation therapy and chemotherapy
16	2016	Ishihara <i>et al.</i> (19)	72	Male	Hypoglycemia	9	Smooth	Surgical resection
17	2016	Mustafa <i>et al.</i> (20)	36	Female	Symptomatic anemia, urinary frequency, increased abdominal girth	6.7×7.5×7.2	Smooth	Surgical resection
18	2016	Ruan <i>et al.</i> (21)	65	Male	Hematuria, urination pain	5.6×4.6×4.0	N/A	Surgical resection
19	2016	Tong <i>et al.</i> (22)	85	Female	Dysuria, urination pain, left abdominal pain	12.3×10.5	Smooth	Surgical resection
20	2017	Kouba <i>et al.</i> (23)	33	Female	Hypoglycemia	3.5	N/A	Transurethral resection
21	2017	Kouba <i>et al.</i> (23)	41	Male	Urinary obstruction	5.7	N/A	Surgical resection
22	2017	Kouba <i>et al.</i> (23)	11	Male	N/A	2.2	N/A	Transurethral resection
23	2017	Tan <i>et al.</i> (24)	76	Male	Urinary frequency	13.5×9.7×21.7	Smooth	Surgical resection
24	2017	Wu <i>et al.</i> (25)	85	Female	Urination pain, dysuria, left lower abdominal pain	10×13×11	Smooth	Partial cystectomy
25	2018	Prunty <i>et al.</i> (26)	49	Male	Abdominal swelling, incomplete bladder emptying, urinary urgency and frequency, thin and loose stools	11	N/A	Surgical resection
26	2019	Rovegno <i>et al.</i> (27)	69	Male	Abdominal pain, hematuria, low urinary tract symptoms	10×8	Not smooth	Radical cystoprostatectomy
27	2019	Cheng <i>et al.</i> (28)	43	Male	Hematuria	8×4.5×3.5	Smooth	Robotic laparoscopic resection of pelvic tumor through bladder
28	2019	Kratiras <i>et al.</i> (29)	31	Male	Frank hematuria, dysuria, dull abdominal pain	4.2×5.3	N/A	Transurethral resection
29	2019	Urbina-Lima <i>et al.</i> (30)	61	Male	Hypoglycemia, generalized asthenia, weight loss	15×23×22	Smooth	Surgical resection
30	2019	Our case	52	Male	Urinary urgency and frequency	7×6.1	Smooth	Partial cystectomy

N/A, not available.

naked eye, tumors showed soft tissue density, clear boundaries and may have cystic areas, calcification, mucoid degeneration or hemorrhage, etc. (34). While in histology, the tumor cells are fusiform and alternately distributed between the dense region (less collagen) and the loose region (abundant collagen) (35). CD34, vimentin, beta-catenin, CD99 and Bcl-2 showed positivity, while CK, EMA, CK-pan, SMA, Calretinin, Desmin, CD56, CD117 or S-100 were lost (26). The CD34 positive expression rate is closely related to the degree of differentiation of tumor cells, it is higher in benign tumors while it decreases in malignant tumors, and is considered to be a specific and essential indicator for the diagnosis of SFT (36). In recent years, some studies have shown that STAT6 can be used as a specific marker of SFT. In this review, all patients could obtain immunohistochemical results showed CD34 positive and 5 of the 7 patients showed STAT6 positive. Malignant SFTs have large tumor size with necrosis and hemorrhage, infiltrative borders, high mitotic count, increased cellularity and cellular pleomorphism (1,26).

Treatment and management

Complete surgical resection is the primary method for the treatment of all SFT. For the patients undergoing surgery, surgical excision with negative margins is the most important factor affecting its prognosis. Chemotherapy can be used for SFT that has been metastasizing or unresectable, but there is currently no clear indication of chemotherapy. The most effective drugs are anthracyclines antibiotic and ifosfamide, supplemented by 2,2-difluorocytosine deoxynucleoside and dacarbazine (37). All the patients in

this review underwent surgical resection, and one patient with recurrence underwent postoperative radiation therapy and chemotherapy. There are only minor complications associated with treatment which is not specific.

The invasiveness and prognosis of SFT are difficult to confirm. Lots of cases in the literature show that the vast majority of SFT manifested as benign, and no recurrence was found after long-term follow-up, still there was potential for metastasis or recurrence (38). In *Table 2*, 1 (1/27, 3.7%) of the 27 patients developed distant metastasis and 5 (5/27,18.5%) presented local recurrence. Unfortunately, due to the lack of data, we are unable to judge the accuracy of risk stratification model for SFT, published by Demicco *et al.* recently, based on the data collected. In the analysis of SFT patients published to date, there was no metastasis in the low-risk group, 7% of the 10-year metastasis risk in the moderate-risk group, and 49% 5-year metastasis risk in the high-risk group (39). Therefore, long-term follow-up is necessary for SFT such as examining abdomen CT every 6 month.

Conclusions

In this paper, we described a rare case of SFT involving bladder and review 29 similar cases in the last decades. The diagnosis of SFT involving bladder should combine clinical presentation and imaging features. Furthermore, cystoscopy and tissue biopsy can assist at times. However, immunohistochemistry is the main proof for the diagnosis of SFT. Complete surgical resection is the primary method, while the method of operation can be open, laparoscopic or robot-assisted. Because of the potential for metastasis or recurrence, long-term follow-up is necessary.

Table 2 Case reports of literature review (postoperative situation)

Case No.	Mitotic count (density-power fields)	Tumor necrosis	Immunohistochemical	Follow-up (months)	Recurrence/metastasis (months)	Complications	Risk stratification model score	Risk class
1	N/A	Present	N/A	48	None/none	None	N/A	N/A
2	N/A	N/A	CD34+, Bcl-2+, CD99+	5	None/none	None	N/A	N/A
3	N/A	N/A	Vimentin+, CD34+/calponin-, S-100-, ALK-, CD117-, CK19-, SMA-	9	None/none	None	N/A	N/A
4	>4/10	Present	CD34+, CD99+/CD117-, SMA-, CKAE1-	18	None/none	None	7	High

Table 2 (Continued)

Table 2 (Continued)

Case No.	Mitotic count (density-power fields)	Tumor necrosis	Immunohistochemical	Follow-up (months)	Recurrence/metastasis (months)	Complications	Risk stratification model score	Risk class
5	0	Absent	CD34+, Bcl-2+	3	None/none	None	1	Low
6	N/A	N/A	CD34+, Bcl-2+/MIB-1<3%	16	None/none	None	N/A	N/A
7	N/A	N/A	CD34+, Bcl-2+, CD99+, vimentin+/CK-, S-100-	41	5 months/none	None	N/A	N/A
8	1/10	Absent	CD34+, Bcl-2+/CKAE1-, CKAE3-, CAM5.2-, SMA-, desmin-, S-100-, CD31-, ALK-1-, CD99-	9	None/none	None	3	Low
9	N/A	N/A	CD34+, STAT6+, Ki-67<2%/ALK-, EMA-, S-100-, 1A4-, HHF-35-, CD117-, CD99-	24	None/none	None	N/A	N/A
10	<1/10	Absent	CD34+, CD99+, C-Kit+, Bcl-2+/CD117-, ALK-1-, SMA-, CK-, S-100-	120	None/none	None	1	Low
11	N/A	Present	CD34+, Bcl-2+, vimentin+, CD99+/SMA-, S-100-, desmin-	24	None/none	N/A	N/A	N/A
12	N/A	N/A	CD34+, Bcl2+, Ki67+/desmin-, S-100-, CK-, A-inhibin-, CD10-, CD68-, ALK-, HMB45-, CD117-	12	None/none	None	N/A	N/A
13	N/A	N/A	CD34+/SMA-, S-100-, desmin-	3	None/none	None	N/A	N/A
14	High mitotic activity	Present	Vimentin+, CD34+, Bcl-2+, beta-catenin+, p53+/CK-pan-, p63-, Calretinin-, SMA-, desmin-, S-100-, CD31-, CD117-, DOG-1-, EMA-, STAT6-, WT-1-	8	None/none	None	6	High
15	High mitotic activity	Present	CD34 +, Bcl-2+, CD99+, vimentin+, Ki-67+/NF-, S-100-, SMA-, CD117-	48	53/none	Slight cystitis	4	Intermediate
16	Low mitotic activity	Present	IGF-II+, CD34+, Bcl-2+, vimentin+/C-Kit-, S-100-	9	144/none	Rectal perforation	3	Low
17	Low mitotic activity	Absent	Vimentin+, CD34+, BCL-2+, CD99+, desmin+, Ki-67 <1% / CKAE1-, CKAE3-, S-100-, CD10-, SMA-, CD117-	N/A	N/A	None	1	Low
18	High mitotic activity	Absent	CD34+, STAT6+, Ki-67+/EMA-, S-100-, CK-, CD117-, desmin-	17	None/none	N/A	4	Intermediate
19	N/A	N/A	CD34+, CD56+/ HMB45-, CD31-, CgA-, CK20-, CK7-, S-100-, SMA-, Syn-	3	None/none	None	N/A	N/A
20	0	N/A	STAT6+, CD34+, Bcl-2+	63	None/none	N/A	N/A	N/A
21	4/10	N/A	STAT6+, CD34+, Bcl-2+	132	72/130	N/A	N/A	N/A
22	6/10	N/A	CD34+/Bcl-2-, STAT6-	12	12/none	N/A	N/A	N/A

Table 2 (Continued)

Table 2 (Continued)

Case No.	Mitotic count (density-power fields)	Tumor necrosis	Immunohistochemical	Follow-up (months)	Recurrence/metastasis (months)	Complications	Risk stratification model score	Risk class
23	1/30	Absent	CD34+, Bcl-2+	12	None/none	Ileus	4	Intermediate
24	0	N/A	CD34+, CD56+/CgA-, CD31-,	N/A	N/A	N/A	N/A	N/A
25	Low mitotic activity	Present	CD34+, Bcl-2+, CD99+, Beta-catenin+/CK-pan-, SMA-, calretinin-, desmin-, CD56-, CD117-, S-100-	3	None/none	None	3	Low
26	Low mitotic activity	Present	CD 34 +, Ki67 =3%/CD117-	N/A	N/A	None	4	Intermediate
27	4/10	Present	Bcl-2+, CD34+, STAT6+, vimentin+, CD99+, Ki-67 =5%/S-100-, EMA-, CK5-, CK-, calretinin-, SMA-, desmin-	3	None/none	None	4	Intermediate
28	10/23	Present	Vimentin+, Bcl-2+, CD99+, CD34+, Ki67 =30-35%/desmin-, CD117-, CKAE1-, CKAE3-	3	None/none	None	4	Intermediate
29	2/50	N/A	CD34+, vimentin+/desmin-, S-100-, CK-, EMA-, CKAE1-, CKAE3-	12	None/none	None	N/A	N/A
30	Low mitotic activity	Absent	CD34+, Bcl-2+, PR+, Ki67 =2%/SMA-, desmin-, DOG-1-, ALK p80-, S-100-, C-Kit-	12	None/none	None	1	Low

CD34, cluster of differentiation 34; Bcl-2, B-cell lymphoma-2; CD99, cluster of differentiation 99; ALK, anaplastic lymphoma kinase; CD117, cluster of differentiation 117; CK19, cytokeratin 19; SMA, smooth muscle actin; CKAE1/CKAE3, cytokeratin AE 1/cytokeratin AE 3; MIB-1, molecular immunology Borstel number 1; CD31, cluster of differentiation 31; STAT6, activator of transcription 6; Ki-67, Ki-67 protein; EMA, epithelial membrane antigen; CD10, cluster of differentiation 10; CD68, cluster of differentiation 68; HMB45, melanoma marker antibody; CK-pan, pan-cytokeratin; WT-1, Wilms tumor 1 protein; IGF-II, insulin-like growth factor II; CgA, chromogranin A; Syn, synaptophysin; CD20, cluster of differentiation 20; CD7, cluster of differentiation 7.

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Footnote

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at <http://dx.doi.org/10.21037/tau.2020.01.09>). The 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. Written informed consent was obtained from the patient for

publication of this manuscript and any accompanying images.

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