REVIEW
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Solitary Fibrous Tumor of the Prostate: What is the Optimal Treatment? Description of A Case and Review of the Pertinent Literature

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Abstract

A solitary fibrous tumor (SFT) originating from the prostate has been rarely reported, presenting the 44th case. We evaluated a 44-year-old man who presented with a two-year history of pressure in the lower abdomen. On magnetic resonance imaging, a 48×66 mm, well-circumscribed mass was observed. 12-core prostatic needle biopsy was performed. Histological examination reported hypocellular and hypercellular areas composed of bland spindle cells arranged in a haphazard pattern. One or two mitotic figures were observed per 10 high-power-fields. Immunohistochemistry analysis showed a strong expression of CD34, STAT-6, and vimentin by tumor cells. We conducted a surveillance protocol for the patient due to the avoidance the surgery. Although there was an increase of approximately 2 cm in tumor diameter, no change was detected in tumor cellularity, number of mitosis, and other histopathological findings in complementary prostatic needle biopsy after three years of follow-up. A literature review of all prostatic SFTs was performed on histopathological features, treatment modality, and reported recurrence and progression data to identify optimal treatment. Local recurrence was reported in five (11.6%) cases and metastasis in two (4.7%) cases. Twenty-two patients underwent radical surgery with a negative margin. None of these had local recurrence and metastasis was reported in only one. Palliative surgery was reported in 18 patients, including five with local recurrence. However, six had no local recurrence or metastasis during the reported follow-up period. Careful surveillance can be conducted in informed patients if there is no malignancy in the histopathologic examination. In all other cases, surgery is strongly advised and should be radical rather than palliative.

Keywords: Solitary fibrous tumor, prostate, immunohistochemistry, STAT6

Introduction

Solitary fibrous tumor (SFT) is a mesenchymal tumor of interstitial dendritic cells (1). Although it was initially considered to be a mesothelioma originating from the pleura, currently, it is reported in many locations due to the widespread presence of dendritic cells outside the thorax. SFTs originating from the prostate have been reported highly infrequently, and this is the 44th case to date.

SFTs are generally benign, although some may show malignant behavior (2). All reported cases have been surgically treated, but there is no standard treatment approach for these very rare tumors, particularly with regard of the benefit of radical surgery. Almost half of the reported cases were treated with palliative surgery, whereas others received radical surgery. Also, surveillance was attempted in one case (3). This report aims to present a rare case and the results of 36 months of surveillance firstly. Secondly, we reviewed all prostatic SFTs in the literature regarding histopathological features, treatment modality, and reported recurrence and progression data to identify the optimal treatment based on the available information.

Case Presentation

Case A 44-year-old man presented with a two-year history of pressure in the lower abdomen. He had no lower urinary tract symptoms (LUTS), or hematuria, and no constipation. He had

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no history of major medical illness. A rectal exam revealed a huge, firm prostate without nodule or induration. His renal function, prostate-specific antigen level, and urine analyses were also within normal limits. On dynamic, gadolinium-enhanced magnetic resonance imaging (MRI) of the abdomen, a 48×66-mm well-circumscribed mass with homogeneous enhancement was observed at the right anterolateral aspect of the prostate (Figure 1a, b). Twelve-core prostatic needle biopsy was performed.

On histological examination of biopsy specimens, the tumor was detected in all six transrectal prostate biopsies of the right prostate lobe and two biopsy specimens from the left lobe. The tumor had hypocellular and hypercellular areas, composed of bland spindle cells arranged in a haphazard pattern. The stroma consisted of a variable number of dense wire-like hyaline collagen deposits, with tumor cells arranged either singly or in small clusters next to the dense collagen (Figure 2a). The spindle-shaped cells had ill-defined borders and scanty eosinophilic cytoplasm. The nuclei were ovoid or elongated, with blunt or tapered ends and contained finely dispersed chromatin or had inconspicuous nucleoli (Figure 2b). Mitotic figures were infrequent. One or two mitotic figures were observed per 10 high-power fields (HPF). No atypical mitotic figure are encountered. These cells did not show prominent atypia or pleomorphism. No lymphovascular invasion or tumor necrosis was observed. Residual prostate parenchyma adjacent to the tumor was noted in some biopsy specimens.

Immunohistochemistry (IHC) analysis showed a strong expression of CD34, STAT-6, and vimentin by tumor cells (Figure 2c, d). Tumor cells were also immunoreactive for CD99, bcl-2, and progesterone (PR) (Figure 3a-c). No staining was observed for CD56, SMA, desmin, pancytokeratin, synaptophysin, CD31, S100, CD117, or DOG1. The proliferation rate, measured by Ki-67 nuclear staining, was evaluated as 5% in hot spots (Figure 3d).

These findings identified an SFT of the prostate. Radical surgery was discussed with the patient. However, he was hesitant about the possible side effects of the surgery, especially urinary incontinence and erectile dysfunction.

The findings were further evaluated according to the malignancy criteria proposed by England et al. (2) and the risk stratification model of Demicco et al. (4) and Pasquali et al. (5). This case had no malignancy criteria as suggested by England et al. (2). Furthermore, the tumor was classified as a low-risk and very low-risk using the models of Demicco et al. (4) and Pasquali et al. (5), respectively. As the biopsy revealed no malignancy and the patient was reluctant to surgery, conservative management was adopted. The tumor was stable on consecutive computed tomography (CT) and MRI scans, three and seven months after diagnosis. However, two years later, CT scan showed that the size of the mass had increased and at that time measured as 77x62x60 mm^3, without any emerging symptoms. Approximately 1 cm additional growth was observed in the tumor (77x82 mm) in the 36th month CT images (Figure 1c, d). Three years after the initial biopsy, confirmatory prostate needle biopsy was performed. According to the histopathological evaluation of the follow-up biopsy, the tumor had the same features as when it was first diagnosed. Histologically, the spindle-shaped tumor cells were dispersed singly or in small groups within the collagenized fibrous stroma. There were no cytological atypia and pleomorphism in follow-up biopsies. The mitotic activity was the same as in the first biopsies. No atypical mitotic figures, tumor necrosis, or lymphovascular invasion were found. These findings indicated that tumor histopathology remained stable during the follow-up (Figure 2e-h).

Literature Review and Discussion

A literature search was conducted in MEDLINE using the following search parameters "(((solitary) AND (fibrous)) AND (tumor)) AND (prostate)." Forty-three cases were identified in 26 reports (3,6-31). Patient age, presenting symptoms, treatment modality, and microscopic findings in terms of malignancy criteria, follow-up time, recurrence, and metastasis information were noted. The cases were evaluated according to the malignancy criteria proposed by England et al. (2) (size >10 cm, mitotic activity >4/10 HPFs, nuclear pleomorphism, infiltrative boundaries, and the presence of necrosis) and divided into two groups (based on the presence of any criterion or none).

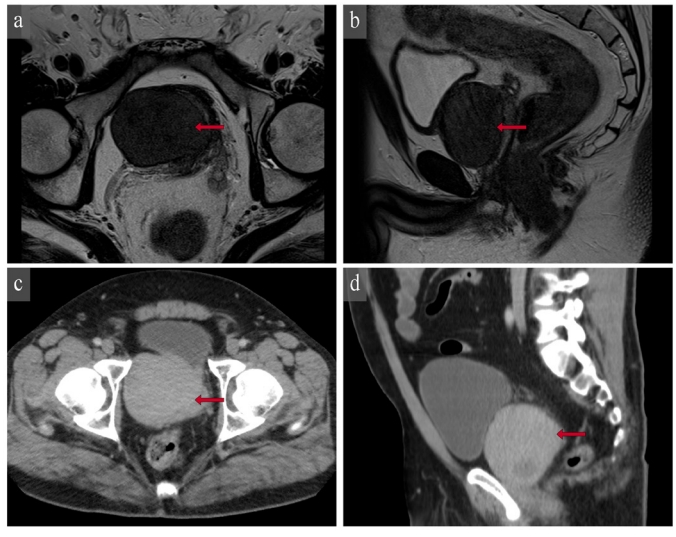

Figure 1. (a) Axial T2WI, **(b)** Sagittal T2WI. On dynamic gadolinium-enhanced magnetic resonance imaging (MRI) of the abdomen, a 48×66 mm well-circumscribed mass and homogeneous enhancement was observed at the right anterolateral side of the prostate. **(c)** Axial CT, **(d)** Sagittal CT images at three years of follow-up. CT scan showed that the size of the mass was increased and measured as 77x82 mm. The arrows indicate the mass

CT: Computed tomography

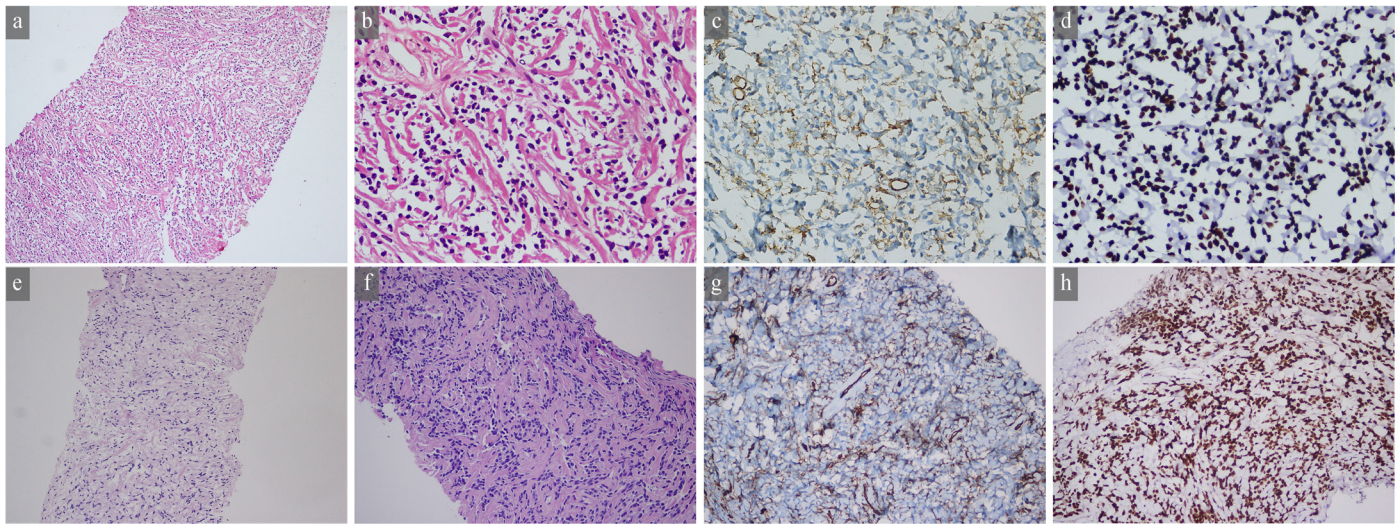

Figure 2. Images from the initial histopathology examination; **(a)** Tumor tissue with haphazard pattern ("patternless pattern") (HEx100). **(b)** High-power view showing oval or elongated nucleus with scant cytoplasm of spindle tumor cells (HEx400). **(c)** Diffuse CD 34 positivity in tumor cells (CD34x400). **(d)** Strong STAT-6 nuclear expression of tumor cells (STAT-6x400). Images of histological assessment of the third-year follow-up biopsy; **(e)** Tumor tissue with haphazard pattern ("patternless pattern") (HEx100). **(f)** High-power view showing oval or elongated nucleus with scant cytoplasm of spindle tumor cells (HEx200). **(g)** Diffuse CD 34 positivity in tumor cells (CD34x200). **(h)** Strong STAT-6 nuclear expression of tumor cells (STAT-6x400)

Demographic characteristics and pathological results of these cases are summarized in Table 1. Most patients were older and suffered from LUTS, with an average tumor size of 8 cm.

Preference of Surgical Procedure

Recurrence and metastasis information was not reported for 15 patients. Local recurrence without metastasis was reported in five (11.6%) patients and metastasis in two (4.65%) patients. Twenty-two (51.2%) patients underwent radical surgery with a negative margin. None of these had local recurrence; metastasis was reported in only one case. Palliative surgery (enucleation or transurethral resection) was reported in 17 (39.5%) patients and radical surgery with a positive margin in one case (6). All the five local recurrences were reported in these cases. However, six of them had no local recurrence or metastasis during the reported follow-up.

Histopathology

Local tumor relapse was reported in five (11.6%) cases without distant metastasis (6,8,15,23,25). Three of them had at least one malignant criterion (mitosis in 7/10 HPFs in one case, nuclear atypia and necrosis in two cases) (6,8,15). The malignant criteria were not clearly specified in the other two cases (23,25). Tumor size was <10 cm in all.

Distant metastasis was reported in two cases (3,6). Both of them had at least one malignant criterion (mitosis in 13/10 HPFs in one case, mitosis present in more than 10/10 HPFs in the other; tumor necrosis in both; hypercellularity in one). Tumor sizes were 6 cm and 9 cm, respectively. Radical surgery with a negative margin was reported in one of them and unspecified in the other.

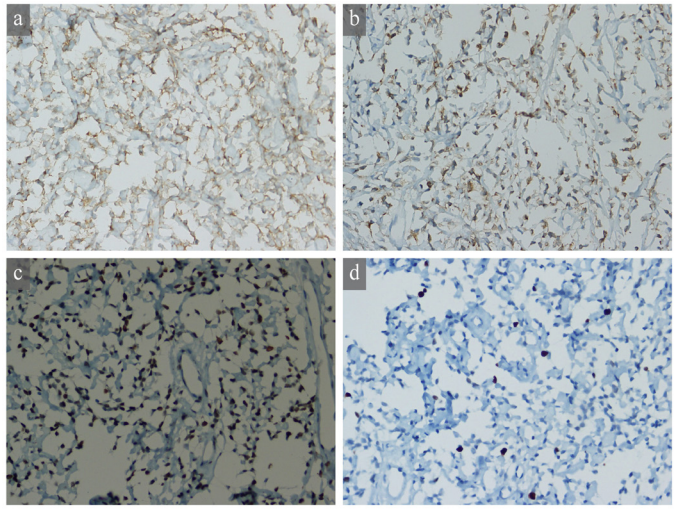

Figure 3. (a) Tumor cells diffusely positive for CD99 (CD99x400). **(b)** Tumor cells diffusely positive for bcl-2 (Bcl-2x400). **(c)** Diffuse, strong progesterone nuclear expression in tumor cells (PRx400). **(d)** Nuclear expression of Ki-67 in some tumor cells (Ki-67x400)

In one of four patients reported by Bakhshwin et al. (6) high-risk SFT by two prognostic systems Salas et al. (32) and Pasquali et al. (5). The patient underwent radical prostatectomy following transurethral resection of the tumor with pT0 disease. However, the patient had a recurrence at the bladder neck and subsequent biopsy-proven metastatic disease to the right obturator lymph node (6). Additionally, Tanaka et al. (3) reported

distant metastasis in their case, although the initial prostatic needle biopsy reported stromal spindle cells with no mitosis. On follow-up MRI approximately ten months later, the mass had increased in size, another prostatic needle biopsy was performed and now showed that tumor cells with round and short spindle-shaped nuclei with some mitoses were present. A total resection of the mass was performed. In the permanent pathological examination, the tumor was found in the muscularis of the prostatic urethra or the bladder. The tumor consisted of spindle cells with fascicular and storiform patterns of growth, and mucinous degeneration and some necrosis were observed in the background. The tumor was hypercellular, and a significant number of mitoses (more than 10/10 HPFs) were present.

Table 1. Summary of demographic characteristics and pathology results of all cases in the literature

	n	**43**
Age	Median (range) years	58 (28-78)
Presentation symptoms		
	Unknown, n (%)	6 (14.0)
	Asymptomatic, n (%)	2 (5.4)
	LUTS, n (%)	27 (73.0)
	Hematuria, n (%)	2 (5.4)
	Urinary retention, n (%)	4 (10.8)
	Constipation, n (%)	2 (5.4)
Tumor diameter		
	Unknown, n (%)	14 (32.5)
	Average (range) cm	8 (1.5-20)
	<10 cm, n (%)	18 (41.9)
	≥10 cm, n (%)	11 (25.6)
Average mitosis/10 HPF		
	Unknown, n (%)	15 (34.9)
	<1, n (%)	15 (34.9)
	1-4, n (%)	5 (11.6)
	≥5, n (%)	8 (18.6)
At least one malignant criterion		
	Unknown, n (%)	6 (14.0)
	Yes, n (%)	19 (44.2)
	No, n (%)	18 (41.9)
Radical surgery with negative margin		
	Unknown, n (%)	4 (9.3)
	Yes, n (%)	21 (48.8)
	No, n (%)	18 (41.9)
Follow-up		
	Unknown, n (%)	15 (34.9)
	Not-specified, n (%)	2 (4.6)
	Median (range) months	18 (2-168)
Local recurrence		
	Unknown, n (%)	15 (34.9)
	Yes, n (%)	5 (11.6)
	No, n (%)	22 (53.4)
Metastasis		
	Unknown, n (%)	15 (34.9)
	Yes, n (%)	2 (4.7)
	No, n (%)	26 (60.5)

HPF: High-power field, LUTS: Lower urinary tract symptoms

In contrast, six cases did not have any tumor recurrence; hence, radical surgery and negative margin were not performed (10,12,19,21,25,30). Median follow-up was 48 (12-168) months in these cases. Three of them had no malignant criterion (10,12,19). However, the others also had at least one malignant criterion (21,25,30). Nair et al. (21) reported a 10 cm tumor that was enucleated with an abdominoperineal approach. They had no evidence of loco-regional recurrence at follow-up after two years. They reported that there was a non-encapsulated tumor on microscopic examination with extended margins containing hyper- and hypo-cellular areas, spindle-shaped with bland nuclei having dispersed chromatin and inconspicuous nucleoli. The mitotic rate was 1/50 HPFs. Pins et al. (25) reported one of two cases, who was treated suprapubic prostatectomy. He had no recurrence after 21 months, though hypercellularity, nuclear atypia, and mitosis 20/50 HPFs were detected in his pathological examination. Xu et al. (30) reported three malignant prostatic SFTs in their study comparing mesenchymal tumors of the prostate. The tumor sizes were 7.6, 19 and 18 cm in largest diameter, respectively. The first was treated with radical prostatectomy with negative margin and excisional biopsies were performed for the others. The last had no information about the follow-up, but the first two cases were followed for six and 84 months without recurrence and metastasis. All tumors had necrosis, and the average mitosis was 5/10 HPFs.

Paraneoplastic Syndromes

On rare occasions SFT can present with paraneoplastic syndromes, the most commonly described being non-islet cell hypoglycemia (33). However, none of the authors reported hypoglycemia in cases of prostatic SFT.

Conclusion

Although there is little data, we suggest that probably the optimal treatment for prostatic SFT is radical surgery with a negative surgical margin. Surgeons should avoid partial resection of the tumor due to the risk of recurrence and metastasis. The malignancy criteria reported by England et al. (2) are a generally useful tool for predicting the prognosis of the disease. However, we did not observe that the tumor diameter affected the results

in the literature. Contrarily, an excessive number of mitosis per HPF seems to be a poor prognostic factor. Surveillance should be performed in patients without malignancy criteria, particularly in cases with very low mitosis rates. Here we report the longest and un-complicated surveillance in the literature. However, one should be careful that insufficient sampling of the tumor with needle biopsies may not show where mitosis is high and nuclear atypia, hypercellularity, or necrosis is present. Close follow-up with repeated biopsy and imaging may be a treatment option in patients younger age and those without malignancy criteria.

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Authorship Contributions

Surgical and Medical Practices: H.Y., A.T.E., Concept: H.Y., A.T.E., Design: H.Y., İ.E.A., Data Collection or Processing: H.Y., İ.E.A., E.Ö., Analysis or Interpretation: H.Y., İ.E.A., Literature Search: H.Y., C.Ö., A.T.E., Writing: H.Y., İ.E.A., C.Ö., E.Ö., A.T.E.

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References

1. Westra WH, Grenko RT, Epstein J. Solitary fibrous tumor of the lower urogenital tract: a report of five cases involving the seminal vesicles, urinary bladder, and prostate. Hum Pathol 2000;31:63-68.
2. England DM, Hochholzer L, McCarthy MJ. Localized benign and malignant fibrous tumors of the pleura. A clinicopathologic review of 223 cases. Am J Surg Pathol 1989;13:640-658.
3. Tanaka Y, Nakamoto A, Inada Y, Narumi Y, Hirose Y, Azuma H. A case of malignant solitary fibrous tumor of the prostatic urethra. BJR Case Rep 2018;4:20180034.
4. Demicco EG, Wagner MJ, Maki RG, Gupta V, Iofin I, Lazar AJ, Wang WL. Risk assessment in solitary fibrous tumors: validation and refinement of a risk stratification model. Mod Pathol 2017;30:1433-1442.
5. Pasquali S, Gronchi A, Strauss D, Bonvalot S, Jeys L, Stacchiotti S, Hayes A, Honore C, Collini P, Renne SL, Alexander N, Grimer RJ, Callegaro D, Sumathi VP, Gourevitch D, Desai A. Resectable extra-pleural and extra-meningeal solitary fibrous tumours: A multi-centre prognostic study. Eur J Surg Oncol 2016;42:1064-1070.
6. Bakhshwin A, Berry RS, Cox RM, Li R, Reynolds JP, Rubin BP, McKenney JK. Malignant solitary fibrous tumour of the prostate: four cases emphasising significant histological and immunophenotypical overlap with sarcomatoid carcinoma. Pathology 2020;52:643-648.
7. Bhargava P, Lee JH, Gupta S, Seyal AR, Vakar-Lopez F, Moshiri M, Dighe MK. Radiologic-pathologic findings of solitary fibrous tumor of the prostate presenting as a large mass with delayed filling-in on MRI. Radiol Case Rep 2012;7:634.
8. Cheng Q, Chang X, Chen W, Qin J, Ai Q, Li H. A rare case of solitary fibrous tumor arising from prostate located inside of bladder. Urol Case Rep 2019;24:100880.
9. Galosi AB, Mazzucchelli R, Scarpelli M, Lopez-Beltran A, Cheng L, Muzzonigro G, Montironi R. Solitary fibrous tumour of the prostate identified on needle biopsy. Eur Urol 2009;56:564-567.
10. Gilbert B, Csillag A, Desai D, McClintock S. Prostate preserving resection of a rare giant peri-prostatic solitary fibrous tumor. Urol Case Rep 2020;32:101167.
11. Herawi M, Epstein JI. Solitary fibrous tumor on needle biopsy and transurethral resection of the prostate: a clinicopathologic study of 13 cases. Am J Surg Pathol 2007;31:870-876.
12. Ishii T, Kuroda K, Nakamura K, Sugiura H. Solitary fibrous tumor of the prostate. Hinyokika kiyo Acta urologica Japonica 2004;50:405-407.
13. Joe BN, Bolaris M, Horvai A, Yeh BM, Coakley FV, Meng MV. Solitary fibrous tumor of the male pelvis: findings at CT with histopathologic correlation. Clin Imaging 2008;32:403-406.
14. Kelly PM, Baxter GM. Solitary fibrous tumour of the prostate. Br J Radiol 1998;71:1086-1088.
15. Liu YT, Song FX, Xiang L, Chang H. Solitary fibrous tumor of the prostate: a case report and 5-year follow-up. Asian J Androl 2019;21:421-422.
16. Manica M, Roscigno M, Naspro R, Sodano M, Milesi L, Gianatti A, Da Pozzo LF. Recurrent retroperitoneal solitary fibrous tumor: a case report and review of the literature. Tumori 2020;300891620974763.
17. Matos J, Paparo F, Calcagno T, Marinaro E, Introini C, Rollandi GA. Solitary Fibrous Tumor of the Prostate. Urology 2020;141:e43-e44.
18. Mentzel T, Bainbridge TC, Katenkamp D. Solitary fibrous tumour: clinicopathological, immunohistochemical, and ultrastructural analysis of 12 cases arising in soft tissues, nasal cavity and nasopharynx, urinary bladder and prostate. Virchows Arch 1997;430:445-453.
19. Mishra A, Corkum MT, Pautler SE, Wehrli B, Winquist E. Images - Solitary fibrous tumor of the prostate. Can Urol Assoc J 2020;14:E613-E614.
20. Moureau-Zabotto L, Chetaille B, Bladou F, Dauvergne PY, Marcy M, Perrot D, Guiramand J, Sarran A, Bertucci F. Solitary fibrous tumor of the prostate: case report and review of the literature. Case Rep Oncol 2012;5:22-29.
21. Nair B, Nambiar A, Hattangadi SB, Sukumar S, Saifuddin MS. Solitary fibrous tumour of prostate: evaluation and management of a rare tumour. Scand J Urol Nephrol 2007;41:442-444.
22. Nishith N, Gupta M, Kaushik N, Sen R. Solitary Fibrous Tumor of the Prostate: A Diagnostic Challenge: A Case Report. Iran J Pathol 2020;15:41-44.
23. Oguro S, Tanimoto A, Jinzaki M, Akita H, Yashiro H, Okuda S, Kuribayashi S, Kameyama K, Mukai M. Imaging findings of solitary fibrous tumor of the prostate: a case report. Magn Reson Imaging 2006;24:673-675.
24. Osamu S, Murasawa H, Imai A, Hatakeyama S, Yoneyama T, Hashimoto Y, Koie T, Ohyama C. Solitary Fibrous Tumor of the Prostate Which Was Initially Misdiagnosed as Prostate Cancer. Case Rep Urol 2017;2017:3594914.
25. Pins MR, Campbell SC, Laskin WB, Steinbronn K, Dalton DP. Solitary fibrous tumor of the prostate a report of 2 cases and review of the literature. Arch Pathol Lab Med 2001;125:274-277.
26. Ronchi A, La Mantia E, Gigantino V, Perdona S, De Sio M, Facchini G, Franco R, De Chiara A. A rare case of malignant solitary fibrous tumor in prostate with review of the literature. Diagn Pathol 2017;12:50.
27. Sekine H, Ohya K, Kojima S, Mizuguchi K. Solitary fibrous tumor of the prostate. Int J Urol 2001;8:137-138.
28. Takeshima Y, Yoneda K, Sanda N, Inai K. Solitary fibrous tumor of the prostate. Pathol Int 1997;47:713-717.
29. Talvitie H, Astrom K, Larsson O, Ahlen J, Bergh A, Egevad L. Solitary fibrous tumor of the prostate: a report of two cases. Pathol Int 2011;61:536-538.
30. Xu Y, Li Z, Shi J, Fu Y, Zhu L, Fan X, Foo WC. Clinicopathological features to distinguish malignant solitary fibrous tumors of the prostate from prostatic stromal tumors. Virchows Arch 2021;478:619-626.
31. Yang W, Sun F, Liu H, Wang G, Shi P, Shao Z, Guo F. Solitary fibrous tumors of the prostate: A case report. Oncol Lett 2015;10:1617-1619.

32. Salas S, Resseguier N, Blay JY, Le Cesne A, Italiano A, Chevreau C, Rosset P, Isambert N, Soulie P, Cupissol D, Delcambre C, Bay JO, Dubray-Longeras P, Krengli M, De Bari B, Villa S, Kaanders J, Torrente S, Pasquier D, Thariat JO, Myroslav L, Sole CV, Dincbas HF, Habboush JY, Zilli T, Dragan T, Khan RK, Ugurluer G, Cena T, Duffaud F, Penel N, Bertucci F, Ranchere-Vince D, Terrier P, Bonvalot S, Macagno N, Lemoine C, Lae M, Coindre JM, Bouvier C. Prediction of local and metastatic recurrence in solitary fibrous tumor: construction of a risk calculator in a multicenter cohort from the French Sarcoma Group (FSG) database. Ann Oncol 2017;28:1979-1987.

33. Robinson LA. Solitary fibrous tumor of the pleura. Cancer Control 2006;13:264-269.