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Solitary Fibrous Tumor of the Prostate: Case Report and Review of the Literature

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Key Words

Prostate · Solitary fibrous tumor · Prognosis

Abstract

Solitary fibrous tumor (SFT), usually described in the pleura, is exceedingly rare in the prostate. We report a 60-year-old man with prostatic SFT revealed by obstructive urinary symptoms, and detected by ultrasonography. Computed tomography (CT) and magnetic resonance imaging suggested a prostatic origin. CT-guided tumor biopsy diagnosed a SFT. A cystoprostatectomy was performed. Pathologic examination showed a 15-cm tumor arising from the prostate and showing histological criteria suggestive of aggressiveness. The surgical resection margins were tumor-free. The patient was then regularly monitored and is still alive in complete remission, 28 months after surgery. In conclusion, we report a new exceptional case of prostatic SFT. We review the literature and discuss the challenging issues of misdiagnosis, prognosis and treatment.

Introduction

Solitary fibrous tumor (SFT) is a rare spindle cell neoplasm most commonly found in the pleura [1]. However, extra-pleural locations have been reported during the last two decades [2]. Their cell of origin and etiology are uncertain, but SFTs likely derive from mesenchymal fibroblastic cells. To our knowledge, the prostatic location is exceptional. It represents a clinical challenge with other spindle cell lesions of the prostate, regarding both the diagnosis and the treatment. Given its scarcity, the continued

documentation of prostatic SFT is important. Here, we report an additional case of prostatic SFT with a 28-month follow-up, and review the literature.

Case Report

In June 2009, a 60-year-old Caucasian man without any specific personal or familial medical history presented with a 1-year history of permanent urinary frequency and urgency. The WHO performance status was 0. His renal function and serum PSA level were normal. He underwent a pelvic ultrasonography (US), which discovered a 13-cm partially cystic well-limited pelvic tumor, located in continuation with the prostate, and associated with a dilatation of the left upper urinary system. Abdo-pelvic computed tomography (CT) revealed a 13-cm heterogeneous pelvic tumor located closely above and likely arising from the prostate, with respect to the fat interface between the mass, the posterior bladder wall and the anterior rectal wall.

The patient was referred to our hospital. Pelvic magnetic resonance imaging (MRI) confirmed the presence of a 10-cm heterogeneous and contrast-enhanced mass, with necrosis inside, causing mass effect on seminal vesicles, with no cleavage plane with the prostate base, associated with a left ureterohydronephrosis. Otherwise, MRI showed fat interface between the mass, the posterior bladder wall and the anterior rectal wall without any sign of involvement ([fig. 1](#)). Histological examination of CT-guided core needle biopsies revealed a spindle cell neoplasm with small, round to fusiform cells strongly expressing CD34 associated with a ramifying vascular network. The suspected diagnosis was SFT. Complete clinical and radiological screening did not detect any lymphadenopathy or distant metastasis. Serum glucose level was normal. The tumor was considered potentially completely resectable. In August 2009, the treatment consisted in a radical cystoprostatectomy with limited bilateral pelvic lymphadenectomy, and vesical reconstruction (Hautmann neobladder). No anterior rectal resection was necessary. Excision was monobloc and macroscopically complete.

Macroscopical examination of the surgical specimen showed a small prostate (4.5 × 4 × 3 cm) and a well-limited, rounded mass (15 × 11.5 × 9 cm) developed from the prostatic base. On cut section, the tumor showed zones of whorled appearance but also cystic, hemorrhagic and necrotic areas. Microscopical analysis ([fig. 2](#)) showed a well-encapsulated lesion composed of spindle cells arranged without any particular architecture (so-called ‘patternless pattern’), associated with variable amounts of collagen and a distinctive vascularization (‘staghorn’ vessels). Part of the tumor was developed within the prostatic base with prostatic glands in contact with tumor cells. Cellular density and mitotic count were very heterogeneous but comprised highly cellular zones and up to 4 mitoses per 10 high-power fields (HPF). By place, fibrosis was predominant and mitotic figures less frequent. There was no capsular effraction, and no vascular embolus. Immunohistochemical (IHC) analysis showed strong expression of CD34 by tumor cells, without expression of epithelial (pancytokeratin, EMA), smooth muscle (alpha-smooth actin), and Schwann cell (PS100) markers. The retained diagnosis was SFT arising from the prostatic base. Moreover, an associated 7-mm prostatic adenocarcinoma was accidentally discovered in the prostatic parenchyma, distant from the capsule, Gleason 6 (3 + 3), without any macro- and microscopical connection with the SFT. The ten removed pelvic lymph nodes were free of tumor.

The postoperative course was uneventful. After surgery, no adjuvant treatment was given, and the patient’s clinical and radiological levels were regularly monitored. A last visit, in November 2011, twenty-eight months after the initial diagnosis, the patient was alive with excellent performance status, without any recurrence of the disease.

Discussion

SFT is rare. It is seen in patients of all ages, but most often between 50 and 70 years [1]. SFT was originally described as a serosa-associated tumor in the thoracic cavity, but in the past 20 years it has been recognized in numerous non-serosal, extrathoracic sites, including the male genitourinary tract. Prostatic SFT is extremely rare. To the

best of our knowledge, only 21 cases have been reported to date in the English literature, as single cases (n = 12) [3–13] and a series of 9 cases [14]. Our case represents the first one with complete clinical, radiological and histological data and a long follow-up.

All cases are summarized in [table 1](#). The median age of patients at the time of diagnosis was 60 years (range 21–87). Clinical symptoms of prostatic SFTs depend on the size (pathological size ranging from 5 to 15 cm) and extension of the tumor: 1 patient was asymptomatic [8], whereas all others presented non-specific urinary and/or rectal symptoms (obstruction, pelvic pressure). One case presented with macroscopical hematuria, which required an emergency hemostatic transvesical prostatectomy. Serum PSA level was normal in all cases tested, and hypoglycemia, previously reported in a few cases of serosal SFT, was not observed in the 13 informative cases. Radiological imaging was useful for the diagnosis and staging, notably to locate the initial site of tumor development, whenever possible. Indeed, these tumors may invade the neighboring structures especially in the retrovesical space, causing difficulty in determining the organ of origin. Generally, the first imaging was US, either transrectal or transabdominal. Pelvic CT and above all MRI more frequently allowed to locate the tumor and to estimate better its locoregional extension before surgery. In our case, and as previously reported [3, 7, 12], MRI findings strongly suggested a tumor origin in the prostate, followed by extension to adjacent organs (especially bladder). The proneness of pleural SFTs to be misdiagnosed is well known [15]. Given the extra-pleural and thus unusual location, prostatic SFTs are even more prone to this problem and represent one of the least commonly found spindle cell tumors on prostatic needle biopsy or transurethral resection. Differential diagnoses are numerous: they include other prostatic benign and malignant tumors, as well as SFTs and other spindle cell tumors arising from adjacent organs (bladder, seminal vesicle, rectum) then invading the prostate, such as rectal GIST. Histological diagnosis of reported cases was provided by analysis of tumor sample, either after needle biopsy (17 cases), transurethral resection (2 cases), or surgical resection without any prior biopsy (4 cases). In all cases, definitive diagnosis was done by histological and IHC examination of the surgical resection specimen. Microscopical and IHC aspects of prostatic SFTs are similar to those of non-prostatic SFTs. In 2 cases (including ours), a small associated prostatic adenocarcinoma was concomitantly accidentally discovered [14].

Regarding the prognosis, 10–20% of pleural SFTs (so-called ‘malignant SFTs’) behave aggressively with local invasiveness and/or recurrences, and/or occasional distant metastases [15, 16]. Histological features used for evaluation of malignancy include large size (>5–10 cm), high mitotic count (>4 per 10 HPF), high cellularity, necrosis, hemorrhage, cytological atypias with pleiomorphism, and infiltrative growth pattern [15, 17]. However, these features do not always forecast an unfavorable clinical outcome. In a large series of pleural SFTs [15], all patients with histologically benign lesions and half of those with malignant lesions (defined by the presence of one or more of the following criteria: cellularity, mitotic count, pleomorphism) were cured by simple excision. Classically, extra-pleural SFTs have a more indolent behavior and are most always defined as benign. However, some of them can display histological aggressiveness criteria and/or present local and/or metastatic relapses [2, 6, 18]. Like SFTs, the clinical outcome of extra-pleural SFTs is unpredictable. This is confirmed for

the prostatic location through the analysis of the 17 cases reported with available follow-up. Only 1 patient displayed a local relapse at 12 months from initial incomplete surgery [12]. Two patients died from postoperative complications on day 1 and from an unrelated cause at 7 months, respectively. None of the 14 other patients did relapse after a follow-up ranging from 2 months to 10 years, although several of them, including ours (large tumor size, cellular areas and foci of necrosis), displayed aggressiveness criteria. The only patient who relapsed was the one with incomplete initial surgery [12]. For many authors, the complete resection of the tumor, whatever its pleural or extra-pleural location, is the most important factor predictive of clinical outcome, implying that the malignant potential of SFTs should be assessed according to both the histological aggressiveness criteria and tumor resectability, keeping in mind that histologically benign and completely resected tumors still have long-term malignant potential. Of course, given the scarcity of reported prostatic SFT cases and the lack of long-term follow-up, no conclusion can be drawn. Additional cases with long-term follow-up are required to better establish prognosis of prostatic SFTs and reliable prognostic criteria.

Given the scarcity of prostatic SFT, data on optimal treatment are obviously limited. Given the possible aggressive behavior and the uncertainty regarding the prognostic factors, prostate SFTs should be removed by complete excision with negative margins, then carefully followed-up for tumor recurrence. Surgery consists generally in a nerve-sparing radical prostatectomy, especially in younger men, aiming at preserving sexual and urinary functions. Associated cystectomy should be reserved to the cases of bladder involvement like ours, where involvement was suspected during surgery. SFTs are relatively insensitive to chemotherapy and radiotherapy [19]. In the literature, one patient received adjuvant radiation therapy after R2 resection.

Conclusion

We report and add an additional case of prostatic SFT to the literature. It is likely that these tumors have a clinical behavior similar to other SFTs. Complete surgical excision with negative margins is the best chance for cure. Although the clinical outcome of prostatic SFT seems favorable, it remains difficult to predict, and postoperative careful long-term follow-up for tumor relapse and possible malignant transformation is warranted regardless the histological criteria of aggressiveness.

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Disclosure Statement

The authors declare no conflict of interest.

Table 1. Twenty-two cases of prostatic SFT reported in the literature

Ref.	Age years	Clinical symptoms	Serum PSA	HypoG	Biopsy	Treatment	Pathol. margins	Pathol. tumor size, cm	Ne-crosis	Mitotic count (per 10 HPF)	Cellularity	Outcome
[6]	72	obstructive urinary S and pelvic pressure	NA	no	no	TUR	NA	NA	no	<2	low	NA
[10]	42	obstructive urinary and rectal S	NA	NA	no	CP	NA	14	yes	NA	NA	NR 10 months
[5]	59	obstructive urinary S	normal	NA	yes	observation	NA	NA	NA	NA	NA	NA
[13]	65*	obstructive urinary S	NA	NA	no	CP + RT	R2	'large'	yes	NA	high	NR 2 months
[8]	57	none (rectal examination)	normal	no	yes	P	R0	10	no	<1	low	NR 15 months
[8]	73	obstructive urinary S	normal	no	no	P	NA	6	no	4	high	NR 21 months
[9]	42	obstructive urinary S	normal	NA	yes	P	R0	NA	NA	2	high	NR 18 months
[4]	21	obstructive urinary S	NA	NA	yes	enucleation (24-mm nodule)	NA	NA	NA	0	NA	NA
[11]	87	obstructive urinary S and hematuria	normal	NA	no	emergency hemostatic P	R1	>9	yes	15	high	Death (day 1 postoperatively)
[12]	35	obstructive urinary S	normal	NA	yes	enucleation	R1	5.2	no	NA	low	Local relapse 12 months
[14]	median: 65** (range 46–75)	obstructive urinary S	NA	no	NA	TUR (1 pt); P (4 pts); CP (2 pts); pelvic exenteration (2 pts); enucleation (1 pt)	NA	median: 10.5** (range 8.5–15)	yes	0 (5 pts); 3–5 (5 pts)	NA	NA (4 pts) death (7 months, unrelated cause: 1 pt) NR 1 to 10 years (5 pts)
[7]	37	obstructive urinary S	NA	NA	yes	enucleation	R1	10	NA	1	high	NR 24 months
[3]	60	obstructive urinary S	normal	NA	yes	P	R0	8	no	1	low	NR 6 months
Our case	60	obstructive urinary S	normal	no	yes	CP	R0	15	yes	4	high	NR 28 months

* This case is also included in [14]; ** 10 cases, including the case reported in [13]. S = Symptoms; NA = not available; HypoG = hypoglycemia; TUR = transurethral resection; CP = cystoprostatectomy; P = prostatectomy; RT = radiotherapy; NR = no relapse.

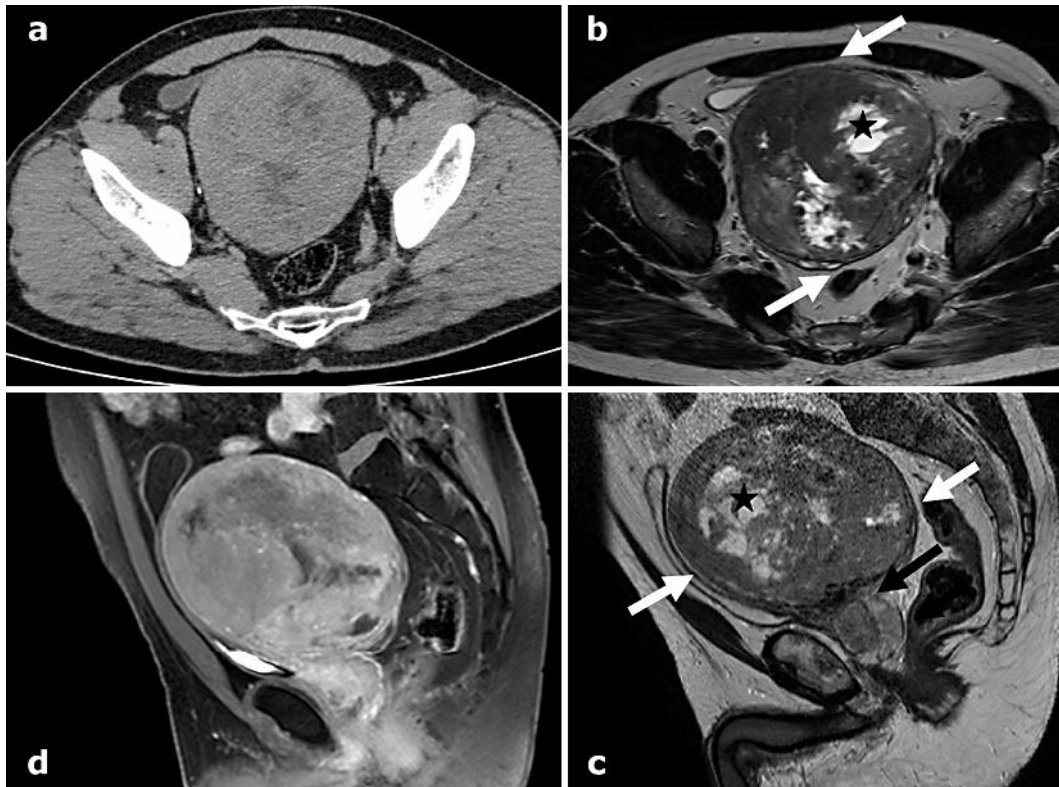


Fig. 1. Imaging of prostatic SFT. Pelvic axial CT-scan (a) detecting a medial massive mass lesion in pelvic and hypogastric regions. Axial (b) and sagittal (c) T2-weighted magnetic resonance image showing an inhomogeneous mass lesion with necrotic areas (stars), arising from prostatic basis (black arrow), with a distinct margin between the mass as well as between the posterior wall of the bladder and the anterior wall of the rectum (white arrows). Sagittal T1-weighted injected magnetic resonance image (d) shows an inhomogeneous and highly contrast-enhanced mass.

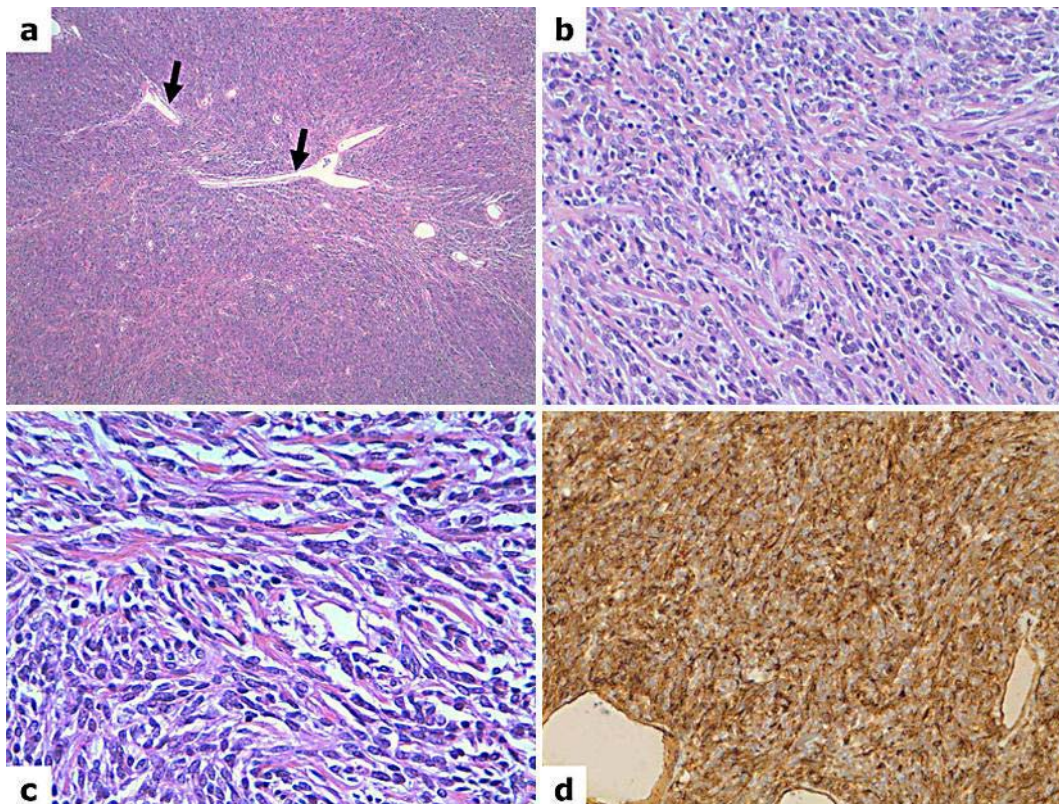


Fig. 2. Prostatic SFT: histological aspects. **a** Low-power view of a highly cellular area with characteristic 'staghorn' vessels (black arrows) (HE staining, original magnification $\times 40$). **b** An area of lower cellularity: cells are mixed with collagen bundles (HE, original magnification $\times 200$). **c** High-power view showing oval to fusiform cells with scant atypias (HE, original magnification $\times 400$). **d** Tumor cells strongly express CD34 (CD34 immunodetection, original magnification $\times 100$).

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