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Original Article

Prostate solitary fibrous tumor: a case report and review of literature

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Abstract: Objective: To investigate the clinical manifestations, imaging features, pathological characteristics and treatment strategies of prostate solitary fibrous tumors. Methods: A retrospective analysis of the clinical data of a male patient with prostate solitary fibrous tumor in our hospital and review of literature. Results: The patient was treated with transurethral resection, followed by confirmation of the disease with immunohistochemistry and pathology. After 3 months, the patient had tumor recurrence and subjected to radical prostatectomy 9 months later. Follow-up for 8 months found that the patient had normal urination with no tumor recurrence. Conclusion: Prostate solitary fibrous tumor is a very rare benign tumor with typical clinical manifestations. Prostate biopsy is the gold standard for diagnosis of the disease. Due to the currently very few clinical case reports, its etiology, pathogenesis, diagnosis and treatment warrant more clinical and basic investigation.

Keywords: Prostate cancer, solitary fibrous tumor, diagnosis, treatment

Introduction

Solitary fibrous tumor (SFT) is a rare tumor originated from mesenchymal spindle cell. SFT was initially thought to occur mainly in the pleura with more common extrapleural SFT than pleural SFT, but accumulating evidence has shown that it happens anywhere in the body, including peritoneal, retroperitoneal space, mediastinum, nasopharynx, eyes, breasts, liver, lung and central nervous system [1]. SFT happens mainly in young adults and the elderly, particularly in women. SFTs consist of diverse benign and malignant tumors. Most SFTs are benign and can be cured by surgery. However, local recurrence or distant metastases occurs in 20% of SFTs patients [2, 3]. Thus it is important to elucidate the pathology and identify biomarkers for the early diagnosis and prognosis of SFTs. Prostate solitary fibrous tumor originates from prostate soft tissues and is extremely rare in clinic. The pathological factors of prostate solitary fibrous tumor remain largely unknown and the currently described pathological factors are sometimes not correlated with the clinical behavior of the tumors [4, 5]. Currently there are no standards for the diagnosis, prognosis, and treatment modality for prostate solitary fibrous tumor. Here we report the diagnosis and treatment of a case of benign prostate solitary fibrous tumor in our hospital.

Patient and methods

General information of the patient

This study was approved by the Ethics Committee of the The First Hospital of Jilin University and written consent was obtained from the patient. This case was a male patient of 53-year old. He experienced dysuria, urgency, lower abdominal and perineal pain 5 years ago. Prostate digital rectal examination (DRE) demonstrated a palpable and hard prostate with size of 5.0×5.0 cm, disappearance of central sulcus, normal ball cavernous muscle reflex, and loss of anal sphincter relaxation, palpable nodules and tenderness. Laboratory tests showed that the serum total PSA was 1.23 ng/ml, and serum free PSA was 0.17 ng/ml. Ultrasound examination detected a 50 mm×40 mm×46 mm prostate with uneven echo; The middle of the prostate significantly enlarged and protruded into the bladder; and there was a 35 mm×36 mm echo mass with

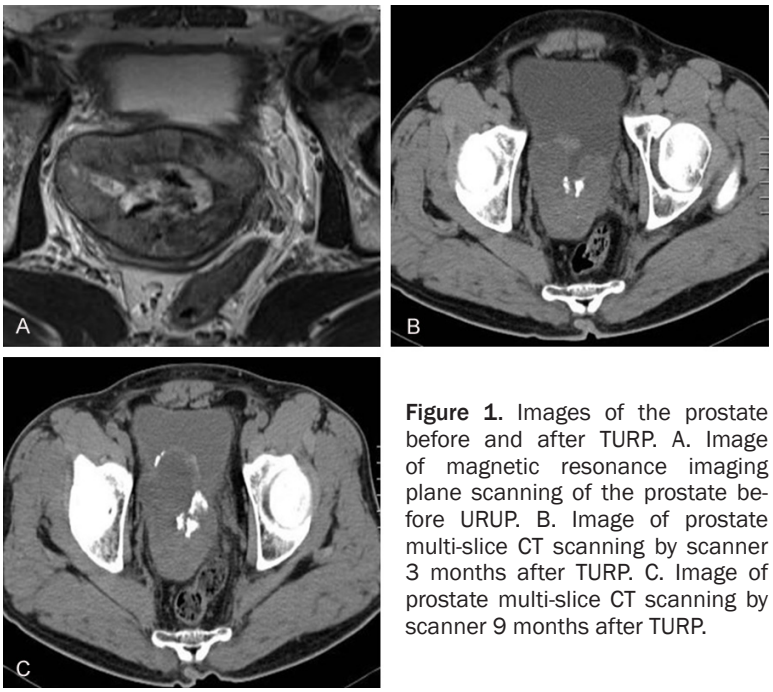

Figure 1. Images of the prostate before and after TURP. A. Image of magnetic resonance imaging plane scanning of the prostate before URUP. B. Image of prostate multi-slice CT scanning by scanner 3 months after TURP. C. Image of prostate multi-slice CT scanning by scanner 9 months after TURP.

unclear border, uneven hyperechoic distribution and no significant blood flow signal. To confirm the diagnosis, prostate tissues were taken by color Doppler ultrasound guided transrectal prostate biopsy and subjected to pathology analyses. The results showed the patient had prostate solitary fibrous tumor. Further immunohistochemistry revealed Bcl-2 (+), CD34 (+), Vimentin (+), CK5 (-), P63 (-), and Actin (-). Patients did not undergo surgery but periodic examination, which showed a progressive increase of the prostate and tumor mass. Fourteen months ago, the patient came to our hospital for re-examination because of increased difficulty in urination. Laboratory tests showed that the serum total and free PSA were 1.9 ng/ml and 0.282 ng/ml, respectively. Urodynamic examination found normal detrusor contractility but bladder outlet obstruction VI level with apparently decreased maximum urinary flow rate. Ultrasonography found the prostate had a size of 77 mm×72 mm×58 mm with partial calcification and uneven echo; no significant change was found in outer gland; however, an echogenic parenchyma tumor with a size of 66 mm×60 mm×54 mm was observed in the internal gland. The tumor had partial calcification with uneven echo and scattered blood flow, and protruded to the bladder with 22 mm. Magnetic resonance imaging (MRI) plane scanning demonstrated enlarged prostate with irregular mass shadow and no clear boundaries with the central and peripheral glands (**Figure 1A**).

Surgery procedures and treatment programs

The patient was treated by transurethral resection (TURP) in our hospital. During the operation, we found the middle of the prostate significantly increased with bilateral lobular hyperplasia and protruded to the bladder thereby pressing urethra, but with no urethral extension; there was calcification within the tumor and a large number of white lime tissues in the glands. The patient was diagnosed as prostate solitary fibrous tumor. Four days after surgery, he recovered to normal urination with no pain, and therefore discharged.

Three months later, the patient came to our hospital for re-examination because of bright red hematuria and dysuria. Prostate multi-slice CT scanning demonstrated an enlarged prostate, inside which an irregular mass shadow of 70 mm 61 mm×64 mm protruding into the bladder was found and demonstrated unclear boundaries with the bladder wall (**Figure 1B**). Laboratory tests showed that the serum total and free PSA were 3.6 ng/ml and 0.598 ng/ml, respectively. Prostate tissues were taken by ultrasound guided transrectal prostate biopsy and subjected to pathology analyses. The results showed the patient had prostate solitary fibrous tumor. Immunohistochemistry results were Ki-67 (+5%), Bcl-2 (+), CD34 (+), CD117 (-), Dog-1 (-), SMA (-), Desmin (-), and S-100 (-).

Six months later, the patient and his family requested radical prostatectomy due to the emergence of intermittent hematuria and aggravated dysuria. Prior to surgery, prostate multi-slice CT scanning was performed and the data demonstrated enlarged and less structured prostate, visible lump cystic mixed density lesions between the prostate and bladder, calcified nodules, unclear boundaries between prostate and bladder rear wall (**Figure**

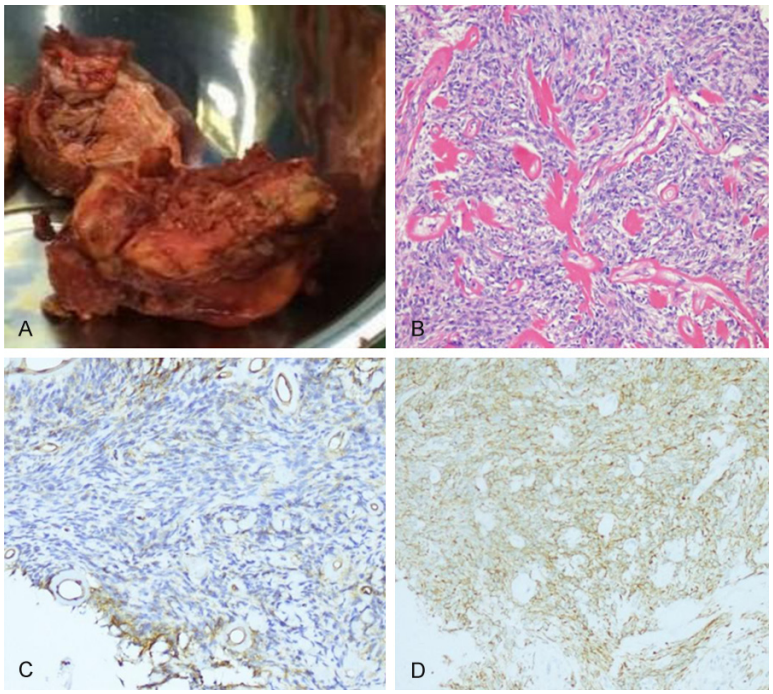

Figure 2. Pathological analysis of the tumor tissue after radical prostatectomy. A. The photo of the removed prostate tumor by surgery. B. The surgical tumor tissues were formalin-fixed and paraffin-embedded. The tissue sections (3-4 μm) were rehydrated and treated with 3% hydrogen peroxide in methanol, followed by staining with hematoxylin and eosin (H&E staining). Magnification: ×20. C. Immunohistochemistry staining of CD34. CD34 was stained brown. Magnification: ×200. D. Immunohistochemistry staining of BCL-2. BCL-2 was positively stained in the diffuse tumor cells. Magnification: ×200.

1C). During the operation, we found the prostate had complete capsule with tough and hard texture and protruded into the bladder about 5 cm; there was calcium deposits in the surface mucosa of the prostate especially the left side; bladder trigone was jacked by the enlarged prostate with normal bilateral ureter. Five days after surgery, he demonstrated no complications of infection, bleeding and organ damages with normal urination, and discharged.

Results and postoperative follow-up

The volume of the tumor removed by radical resection was 80 mm×75 mm×50 mm. No tumor invasion was found in the bladder, urethral margin, prostate outer circumferential margin, vas deferens margin, and seminal vesicles (**Figure 2A**). Pathological analysis showed it was a solitary fibrous tumor with ill-defined local boundary and aggressive growth; there was a large area of necrosis, infarction and scattered calcification; there was increased focal cell density, atypia and mitotic nuclei (**Figure 2B**). Immunohistochemistry analysis showed that tumor tissues were positively stained for CD34 in local tumor foci (**Figure 2C**), and positively stained for Bcl-2 throughout the tumor tissues (**Figure 2D**). The patient was followed up for 8 months and showed normal urination, disappearance of pain and hematuria, and no tumor recurrence and metastasis.

Discussion

SFT is a rare spindle cell soft tissue tumor and is generally believed to originate from CD34-positive dendritic interstitial cells. SFT mainly occurs in the chest visceral pleura with varying morphology. Benign SFT is an isolated tumor mass with clear boundary and fake fiber capsule and shows expansive growth; while malignant SFT has an undefined boundary with invasive growth. SFT has been rarely found in other organs, particularly prostate [6]. Prostate solitary fibrous tumor was first discovered and named in 1931 by Klemperer et al [7]. Up to now, together with the present report, only 36 case of prostate solitary fibrous tumor have been reported worldwide [6, 8-19]. Cancer results from multiple genetic and epigenetic alterations through a period of process [20]. However, due to the very limited cases, the molecular markers for the diagnosis and prognosis of prostate solitary fibrous tumor remain elusive. Moreover, the development of targeted therapy for malignant prostate solitary fibrous tumor await for the identification and validation of the driver genetic mutations for prostate solitary fibrous tumor.

Clinical manifestations of prostate solitary fibrous tumor

The reported prostate solitary fibrous tumor patients were 33-78 years (mean 55 years)

with tumor diameter of 4.7-25 cm (average of 9.8 cm). In the reported fourteen cases from China, all demonstrated symptoms of dysuria, 7 cases of frequent urinarination and urgency, 4 cases of dysuria, 3 cases of constipation, 2 cases of hematuria; only one case had abnormal PSA; pathological analysis found 8 cases of benign SFT and 6 cases of malignant SFT; immunohistochemistry showed that, all were CD34 positive, 8 cases were Vim and bcl-2-positive, six cases were CD99-positive, and six cases were SAM and S-100 negative [9-19]. Early prostate SFT often demonstrates no obvious clinical symptoms; however with the increase of the tumor volume, patients show apparent clinical manifestations such as progressive difficulty in urination, frequent urination, urgency, dysuria, hematuria, constipation and abdominal pain.

Imaging features and laboratory tests of prostate solitary fibrous tumor

B-ultrasound images of prostate solitary fibrous tumor demonstrate a significantly enlarged prostate with echo mass inside; clear boundary and evenly distribution is often associated with no blood flow signals, indicating a benign tumor; while less clear boundary and uneven distribution is frequently accompanied with blood flow signals, suggesting malignancy. Either CT or MRI can clearly show the prostate size, shape, and density, and whether the boundary is clear, whether there is invasive growth, whether the prostate presses forward bladder, and whether oppresses rectum and sigmoid colon backward. Typical CT image of prostate solitary fibrous tumor is a mass of low density. MRI imaging features are T1-weighted low density mass and T2-weighted high-density mass [1, 21]. When the above imaging features are observed, the possibility of prostate solitary fibrous tumor is very high and prostate biopsy to confirm the diagnosis should be considered as early as possible. Patients with prostate solitary fibrous tumor have normal range of PSA and other serological tests are also normal. Therefore, PSA level is not considered as an SFT screening indicator.

Diagnosis of prostate solitary fibrous tumor

It was proposed that malignant pleural SFT might have the following conditions: rich tumor cells are densely arranged; mitotic counts are more than 4/10HPF; cells show atypia; there are no tumor boundaries and pedicles; there is a lot of tumor necrosis; tumor size is more than 10 cm [14]. Most pathologists believe that the morphological characteristics of pleural SFT are equally applicable to the judgment of extrapleural SFT. Sun et al [22] reported that immunohistochemical analysis of basic fibroblast growth factor and Ki-67 expression can distinguish between benign and malignant SFT. Pathological analysis of prostate biopsy is the gold standard for diagnosis of prostate solitary fibrous tumor. The microscopic features of prostate biopsy include: (A) Tumor realm is clear or coated with a pseudo fibrous capsule; there is infiltration around the tumor in malignant tumors; tumor cells are short spindle, round or oval with less or unclear cytoplasm; tumor cells show uniform nuclear chromatin and unclear nucleolus; Mitotic counts range from 0 to 4/10 high-power fields (HPF) with large than these mitotic counts in malignant tumor. (B) Tumor mass consists of alternating cell rich region and collagen fibers composed of cell sparse areas; in cell sparse area, the collagen fibers are thick and rope-like bundle, sometimes presented as scar-like or unstructured solitary fibrous tumor. (C) The arrangement of tumor cells is various, including disorganized structure, fence-like hemangiopericytoma, wavy and phyllodes tumor [9].

Treatment of prostate solitary fibrous tumor

Due to the limited cases, treatment standard for prostate SFT has not yet been established. Radical prostatectomy and transurethral resection have been applied for the treatment of prostate SFT. In china, 8 cases were treated with radical prostatectomy, and follow up found 3 cases of recurrence; 4 cases were treated with transurethral resection, and follow up observed 2 cases of recurrence. To improve the understanding of the pathogenesis of the disease and find effective treatment strategies, long-term follow-up should be performed for these patients. Moreover, numerous targeted therapies have been developed for other common tumors such as lung cancer and breast cancer. Nevertheless, molecular targeted therapy for prostate solitary fibrous tumor has not yet been developed. It is thus important to find the genetic drivers of the tumorigenesis and thereby identify the targets for the develop-

ment of molecular targeted therapy for prostate solitary fibrous tumor.

In summary, prostate solitary fibrous tumor is an extremely rare tumor in clinical practice and demonstrates typical lower urinary tract obstruction. At present, the relevant literature is very limited. In order to improve diagnosis and find effective treatment modalities for prostate solitary fibrous tumor, more and in-depth studies are needed to elucidate the etiology and pathogenesis. Particularly, with the rapid advance of next-generation sequencing, precision medicine will be the future trend for the diagnosis and treatment of cancers. It will be important to perform whole genome and RNA sequencing of prostate solitary fibrous tumors and find the biomarkers for the diagnosis, prognosis and treatment of prostate solitary fibrous tumor.

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Disclosure of conflict of interest

None.

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