Prostate Biopsy in Young Patients with Hematospermia Without an Indication for Biopsy Contributed to the Early Diagnosis of Prostate Sarcoma: A Case Report

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Abstract

Background: Prostate sarcoma is a rare group of prostate primary malignant tumors with a poor prognosis, and its early diagnosis is challenging. Here we report a case of early diagnosis of prostate sarcoma and summarize the clinical characteristics of this group of patients.

Case presentation: We present a case of a 40-year-old male patient with prostate leiomyosarcoma having hematospermia as the initial symptom. His prostate-specific antigen was 2.97 ng/ml, magnetic resonance imaging showed a prostate nodule with possible bleeding at the lesion margins with Prostate Imaging Reporting and Data System (PI-RADS) category 2. According to the guidelines, prostate biopsy was not recommended, but we learned from our previous experience and still recommended him for prostate biopsy. With his consent, a combined prostate biopsy was performed, and he received an early diagnosis of prostate leiomyosarcoma 19 days after the initial symptom of hematospermia. Reviewing previous cases of prostate sarcoma patients at our medical center, we found that the best time for diagnosis and treatment were generally missed.

Conclusions: Combination with magnetic resonance imaging is contributive to the early diagnoses of prostate sarcoma. Therefore, when young patients present with repeated hematospermia, even if they have normal prostate-specific antigen, it is important to be alert to the possibility of prostate sarcoma, and multiparametric magnetic resonance imaging rather than ultrasound is recommended, and if MRI reveals prostate lesions, it is recommended to be biopsied regardless of PI-RADS categories, and a combined biopsy is recommended.

Introduction

Prostate sarcoma is a rare type of prostate primary malignant tumors with a rapid progression and poor prognosis(1). Early diagnosis and early treatment of prostate sarcoma improve the prognosis of patients, however, early diagnosis is challenging(2). The difficulty in early diagnosis is mainly due to the rareness of prostate sarcoma and the fact that it tends to occur in young men, which is very different from prostate cancer. When these young patients show symptoms such as dysuria, frequent urination, urgent urination, painful urination, and hematuria, an ultrasound examination is generally recommended. However, an early-stage prostate sarcoma can be insignificant on ultrasound and can be misdiagnosed as seminal vesiculitis or prostatitis. Later when the patients' symptoms worsen, they will go back to the hospital for further examination, and by then the best time for treatment is often missed.

Prostate Imaging Reporting and Data System (PI-RADS) is an analysis system for multiparametric magnetic resonance imaging (mpMRI) results of prostate cancer(3). Only 3% of PI-RADS categories 1–2 lesions are diagnosed as clinically significant prostate cancer (csPCa) by systematic biopsy(4), and therefore prostate biopsy is not recommended for PI-RADS categories 1–2 according to the guidelines, unless other clinical risk factors are considered(5, 6). Instead, in our case report, the patient with PRADS category 2 and normal PSA level was found to have prostate leiomyosarcoma.

We believe that at least MRI should be performed in patients with repeated hematospermia instead of ultrasound even if they had normal PSA level to detect an early-stage prostate sarcoma, and if there are any suspicious findings, regardless of PI-RADS categories, prostate biopsy should be performed.

Case Presentation

A 40-year-old male presented to our urology department with hematospermia. He had continuous hematospermia after several sexual intercourses for the last one month. The hematospermia symptom was not accompanied by painful ejaculation, painful discomfort in the genitals, hematuria, scrotal enlargement, bleeding from other parts of the body, skin petechiae, hot flashes or night sweats. He had chronic symptoms of frequent urination and urgent urination and he was diagnosed with benign prostatic hyperplasia 8 years ago and did not take treatment. His vital signs were stable. A slightly

enlarged prostate with a shallow central sulcus and a tougher texture were revealed in the rectal examination, no nodules were found, no pressure pain and the rest of the physical examination was unremarkable. He underwent a PSA and MRI examination. The results showed that TPSA was 2.97 ng/ml, FPSA was 0.249 ng/ml, F/T was 0.08. A nodule was present on the left peripheral side of the prostate, with possible bleeding at the margins of the lesion; besides, the prostate was mildly enlarged, volume 50*40*43 mm, and PI-RADS assessment category was 2, meaning the presence of clinically significant prostate cancer is unlikely (Fig. 1A-L). According to the guidelines(6), prostate biopsy is not recommended, but we learned from our previous experience and still recommended him for prostate biopsy. With the patient's consent, he underwent a combined biopsy (Fig. 2). Immunohistochemical results showed CK (-), EMA individual cells (+), CD34 (-), MyoD1 (-), CgA (-), Syn (-), NSE (-), Myogenin (-), desmin (+), SMA (+), Caldesmon (+), Vimentin (+), ki67 60% (+). The pathological results showed that the left lobe biopsy result was malignant, and combined with immunohistochemistry and histomorphology, it was consistent with leiomyosarcoma.

Based on the pathological findings and the MRI results, the diagnosis of prostate leiomyosarcoma could be confirmed.

The patient underwent a further PET/CT examination to clarify the tumor's metastasis. The PET/CT whole-body imaging results showed a low-density mass in the left outer zone of the prostate, measuring approximately 3.0*2.7 cm, with PET showing a heterogeneous increase in radioactive uptake and a SUV_{max} value of 6.6, which was consistent with the characteristics of leiomyosarcoma. In addition, the punctate dense shadow was seen in the prostate. The prostate was enlarged with calcification. No abnormalities were detected in the bladder, bilateral seminal vesicles, or rectum, and no distant metastases were detected (Fig. 1M).

He was subsequently treated with robotic-assisted laparoscopic prostatectomy. After surgery, his condition was stable and he had no adverse reactions. After 7 days of observation, he felt well and was discharged from the hospital. Postoperative pathological findings were consistent with biopsy results (Fig. 3). Postoperative pathology showed no tumor involvement was found in the prostate basal and apical margins, which indicated that he might have a better prognosis. Three months after surgery, the patient's follow-up MRI results showed no recurrence or metastasis. His medical timeline is present in Fig. 4A.

From 2012 to the present, we have admitted five patients with prostate sarcoma, three with leiomyosarcoma and two with rhabdomyosarcoma (Table 1). We found that PSA were normal in all five patients. And their age distribution is wide, ranging from 26 to 80, with a median age of 40. Their duration from initial symptoms to diagnosis differ from 19 days to over 1 year. With the exception of the case we reported, other 4 prostate sarcoma patients all died of distant metastases, with overall survival range from 3 months to 7 months. We summarized their treatment process and found that most prostate sarcoma patients were missed at their first visits (Fig. 4).

Table 1
Clinical Characteristics of 5 prostate sarcoma patients at our hospital.

Patient	PT001	PT002	PT003	PT004	PT005
Age	40	50	26	28	80
Initial symptoms	hematospermia	hematuria	dysuria and hematospermia	frequent urination, urgent urination, painful urination	hematuria
PSA (ng/ml)	2.97	2.88	1	2.31	3.10
US suggestive of a tumor when diagnosed	(-)	(+)	(+)	(+)	(+)
MRI suggestive of a tumor when diagnosed	(-)	(+)	(+)	(+)	(+)
How to get diagnosed	Combined Biopsy	Surgery	Surgery	TRUS	TRUS
Diagnosis	leiomyosarcoma	leiomyosarcoma	leiomyosarcoma	rhabdomyosarcoma	rhabdomyosarcoma
Duration from initial symptoms to diagnosis	19 days	14 months	12 months	10 months	12 months
Recurrence and metastasis in three months after surgery	(-)	(+)	(+)	(+)	(+)
Overall survival	NA	7 months	3 months	4 months	3 months
Invasion and metastasis on diagnosis					
Basement membrane	(-)	(+)	(+)	(+)	(+)
Seminal vesicles	(-)	(-)	(+)	(-)	(-)
Vas deferens	(-)	(-)	(-)	(-)	(+)
Lymph Node	(-)	(-)	(+)	(+)	(-)
Bladder	(-)	(+)	(+)	(-)	(+)

Patient PT001 is the patient described in the text, other 4 are previous patients at our medical center. Abbreviation: PSA, prostate-specific antigen; US, ultrasound; MRI, magnetic resonance imaging; TRUS, transrectal ultrasound-guided prostate biopsy.

Patient	PT001	PT002	PT003	PT004	PT005
Rectum	(-)	(-)	(+)	(+)	(-)
Lung	(-)	(+)	(+)	(+)	(-)
Liver	(-)	(-)	(+)	(-)	(-)
Bone	(-)	(-)	(-)	(+)	(+)
Patient PT001 is the patient described in the text, other 4 are previous patients at our medical center. Abbreviation: PSA, prostate-specific antigen; US, ultrasound; MRI, magnetic resonance imaging; TRUS, transrectal ultrasound-guided prostate biopsy.					

Discussion

Prostate sarcoma is very rare, with an incidence of less than 0.1% of prostate malignancies(1). It has a poor prognosis and relies primarily on surgical treatment. Generally, prostate sarcomas extend beyond the prostate and invade adjacent structures(7). Patients who do not have invasive cut margins, basement membrane invasion, prostate apex involvement, or distant metastases tend to have a better prognosis(8, 9). The tumor of the patient reported above did not break through the basement membrane and we believe that he would have a better prognosis. Therefore, early diagnosis of the disease and prompt surgical treatment will greatly improve the prognosis. Our case of a patient whose diagnosis was confirmed by early biopsy beyond the guidelines provides a precedent for future clinical work. Prostate biopsy is recommended for the early detection of prostate sarcoma in young patients with recurrent haemospermia, even if their PSA is not high and with PI-RADS categories 1–2, and a combined biopsy is recommended for its high diagnostic accuracy compared with targeted biopsy alone or systematic biopsy alone(10).

Referring to all 5 patients with prostate sarcoma at our medical center from 2012 to the present, we found that the duration between their initial symptoms and diagnoses varied, mainly because patients visited the hospital for symptoms such as dysuria, hematuria and hematospermia, they underwent ultrasound but ultrasound did not reveal the possibility of an early-stage prostate sarcoma, so they were considered to have seminal vesiculitis or prostatitis and were treated accordingly. As the tumor progressed, symptoms would worsen, and the patient would come for further examination. However, it was the general situation that the prostate sarcoma has extended beyond prostate or has even formed distant metastasis(7). Therefore, a detection method combined with MRI is necessary to improve patients' prognoses, and MRI rather than ultrasound is recommended for young patients with repeated hematispermia and normal PSA. Besides, we found that PSA was difficult to have positive findings, even in patients who have developed invasion and metastasis to adjacent organs(Table 1), which is consistent with the previous findings(7). Their overall survival was relatively short compared to other reported patients (8, 11), mainly because these patients had metastatic tumor at the time of diagnosis and subsequently had positive surgical margins, which indicated poor prognoses.

The present study has limitations. There were not enough prostate sarcoma samples, and large-scale clinical trials are required to prove the necessity for young patients with repeated hematospermia to undergo MRI and combined prostate biopsy to avoid worse clinical outcomes.

Conclusion

When young patients present with repeated hematospermia, even if they have normal PSA, multiparametric magnetic resonance imaging (mpMRI) is recommended, and if MRI results show a lesion, it is recommended to be biopsied regardless of PI-RADS categories, and a combined biopsy is recommended.

Abbreviations

PSA	Prostate-Specific Antigen
US	Ultrasound
PI-RADS	Prostate Imaging Reporting and Data System
MRI	Magnetic Resonance Imaging
mpMRI	multiparametric magnetic resonance imaging
csPCa	clinically significant prostate cancer
TRUS	Transrectal Ultrasound-guided prostate biopsy

Declarations

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Authors' contribution

Fei Gao designed this study. Fei Gao and Zhuoyuan Yu had full access to all data in the study and take responsibility for the integrity and accuracy of the data. Zhuoyuan Yu, Fei Gao, Mei Yang and Xiaoyan Liu drafted the manuscript. All authors collected the data. All authors have revised and approved the final manuscript.

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Availability of data and materials

Not applicable.

Declarations

Ethics approval and consent to participate

Not applicable.

Consent for publication

Written informed consent was obtained from the patient for the publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Competing interests

The authors declare that they have no competing interests.

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Figures

Figure 1

Magnetic resonance imaging (MRI) and PET/CT images of the patient. A, E, I: Sagittal section of the T2 image of the pelvic cavity. B, F, J: Cross-section of the T1 image of the pelvic cavity. C, G, K: Cross-section of the T2 image of the pelvic cavity. D, H, L: Cross-section of the DWI image of the pelvic cavity. M: PET/CT images of the patient. The green circle indicated the abnormal area of the prostate.

Figure 2

Images of combined prostate biopsy. A: The MRI image of the patient. The red and green circle indicated abnormal area in the MRI image. B: The ultrasound image of the patient. The arrow indicated the biopsy needle.

Figure 3

Hematoxylin and eosin (H&E) staining of the patient's prostate. A: HE staining of prostate leiomyosarcoma. B: HE staining of normal prostate tissue. (200x, scale bar 100um).

Figure 4

Timeline of 5 prostate sarcoma patients at our medical center. The red bars stand for the duration between the initial symptoms and diagnoses. The green bars represent the overall survival of the patient from the time of diagnosis. A: Timeline of patient PT001, he was diagnosed 19 days after initial symptoms. B: Timeline of patient PT002, he was diagnosed 14 months after his initial symptoms. C: Timeline of patient PT003, he was diagnosed 2 months after his initial symptoms. D: Timeline of patient PT004, he was diagnosed 3 months after his initial symptoms. E: Timeline of patient PT005, he was diagnosed 2 months after his initial symptoms. Abbreviation: OS, overall survival; MRI, magnetic resonance imaging; PSA, prostate-specific antigen; US, ultrasound; ICU, intensive care unit; TRUS, transrectal ultrasound-guided prostate biopsy.