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### **Case Report**

# Prostate leiomyosarcoma treatment using three-dimensional reconstruction: a case report and literature review

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### Abstract

**Background**: Prostate sarcoma is a malignant tumor from the prostate stroma. However, its pathogenesis is unknown. This is a rare type of prostate tumor.

**Case Presentation**: A rare case of prostate leiomyosarcoma has been described. A 55-year-old prostate leiomyosarcoma patient who suffered from progressive dysuria and frequent urination for half a year was examined. Pathology, immunohistochemical staining, and laparoscopic radical prostatectomy were performed using three-dimensional (3D) reconstruction to diagnose prostate leiomyosarcoma. The patient did not receive adjuvant therapy after the operation. Final pathology was used to confirm prostate leiomyosarcoma. After one year of follow-up, the patient regularly underwent a digital rectal examination and abdominal MRI. However, no tumor recurrence or metastasis was found.

**Discussion**: We discussed diagnosis and treatment of prostate leiomyosarcoma and reviewd literatures.

**Conclusion**: Prostate leiomyosarcoma is a rare special type of prostate tumor, and its diagnosis is mainly based on pathological features. Radical excision is the primary treatment for prostate leiomyosarcoma. The 3D reconstruction is essential in the surgery because it can determine the size of the tumor and its relationship to the surrounding tissue. A postoperative adjuvant therapy can also be performed.

### Keywords

Prostate leiomyosarcoma; diagnosis; treatment; three-dimensional reconstruction; case report

### 1. Introduction

Prostate sarcoma is a malignant tumor from the prostate stroma, with a low incidence of 0.1% of all prostate tumors [1, 2]. Leiomyosarcoma accounts for 25% of prostate sar-

comas. Prostate leiomyosarcoma is a severe tumor with high malignancy and poor prognosis. Its treatment is mainly through surgical resection combined with radiotherapy and chemotherapy. In the following case, the radical operation





FIG. 1. Prostate MRI: prostate was enlarged with abnormal shape, about 6.3 \* 5.2 cm in size, significantly heterogeneous enhancement in enhanced scan. The local and seminal vesicle boundary was not clear. There was no thickening in the lower segment of bilateral ureter and the tube wall. There was no enlarged lymph node in the pelvic cavity. A diagnosis of prostate cancer.

was performed using 3D reconstruction to provide a reference for prostate leiomyosarcoma diagnosis and treatment. The case is reported following the CARE guideline for case reports [3]. 3D printing was developed by Chuck Hull and was initially reported in the early 1980s [4]. Since its invention, this technology has become an integral tool in the rapid prototyping and manufacturing of many items, such as clothes, furniture, houses, cars, aircraft, and weaponry [5-9]. The development of 3D printing technology and computer technology makes it possible for physicians and surgeons to turn traditional medical imaging technologies (such as computed tomography (CT) and magnetic resonance imaging (MRI)) into high-resolution, patient-specific, 3D models of complex anatomy. This process is called 3D reconstruction which refers to the process of reconstruction of 3D information from single or multi-view images.

#### 2. Case presentation

A 55-years-old man with a 6-month history of progressive dysuria and frequent urination visited a hospital on July 17, 2019. He was a farmer with no family history of the malignancy and had no history of diabetes, hypertension, or tuberculosis. He had no significant surgical history or treatment in the past.

Digital rectal examination identified the prostate as enlarged and tougher, with a smooth surface and no central sulcus. Besides, there were no nodules and touching tenderness. Laboratory examination showed that there were no known abnormalities in urine and stool routines. Prostatespecific antigen (PSA), neuron-specific enolase (NSE), and tumor-associated material (TAM) concentrations were 0.737 ng/mL, 72.18 ng/mL, and 112 U/mL (normal 0-95 U/mL in our hospital), respectively. Other tumor indicators were normal.

Prostate ultrasound showed that prostate volume significantly increased (5.9 \* 6.0 \* 6.0 cm) with full shape and uneven parenchymal echo, indicating strong speckled echo. Prostate MRI (A 3.0T MR system. Images were acquired during free breathing. The anatomical MRI was performed using fast spin echo (FSE) sequence to acquire T1-weighted images in the axial, coronal, and sagittal plane with the following imaging parameters: repetition/echo time (TR/TE) 560/7.176 msec, number of excitations (NEX) 2, slice thickness of 2 mm, space 0.5 mm, and T2-weighted images in the axial, coronal, and sagittal planes, with the following parameters: TR/TE 5600/105.7 msec, NEX 2, slice thickness of 5 mm, space 0.5 mm. DWI was acquired using a singleshot echo-planar sequence (SSEPI) with 12 b values (0, 25, 50, 75, 100, 150, 200, 400, 600, 800, 1000, 1200 s/mm<sup>2</sup>), TR/TE of 3820/95 msec, field of view (FOV) 340 \* 340 mm, matrix 128 \* 160, slice thickness 5 mm, space 0.5 mm, bandwidth 250 kHz, NEX 2, 2, 2, 3, 3, 4, 4, 6, 6, 6, 8, 8, corresponding with each b value. The acquisition time of DWI with ultrahigh b-values was 6 minutes and 56 seconds.) showed that the prostate was enlarged with an abnormal shape of about 6.3 \* 5.2 cm (in the axial plane), and was significantly heterogeneous in the enhanced scan. However, the local and seminal vesicle boundary was not clear. There was no thickening in the lower segment between the bilateral ureter and the tube wall and no enlarged lymph node in the pelvic cavity. Prostate cancer diagnosis should be considered (Fig. 1).

Pathology of prostate biopsy demonstrated that the biopsy tissues with focal necrosis had a heterotypic spindle, and the cell nucleus was divided. Together with the immunohisto-



FIG. 2. Prostate biopsy pathology: Haematoxylin and Eosin staining with ×40 magnification demonstrating the biopsy tissues with focal necrosis showed heterotypic fusiform, and the cell nucleus were divided. Combined with the immunohistochemical results, the lesions were consistent with leiomyosarcoma.



FIG. 3. Expression of the prostate tumor markers. (A) CD34. (B) Desmin. (C) Ki-67. (D) P63. (E) PSA. (F) S100. (G) SMA. (H) Vim.

chemical results, the lesions were consistent with leiomyosarcoma (Fig. 2A). Immunohistochemical stains revealed that neoplastic cells were strongly positive for desmin and Smooth Muscle Actin (SMA), focally positive for Vimentin (Vim) and Cytokeratin (CK), and negative for P504S, PSA, myogenin, CD34, s-100, P63. The Ki-67 index was 70%, indicating nuclear reactivity (Fig. 3). A 3D reconstruction software (ONIS 2.5) was used to transform multiple views of the MR into a 3D virtual model showing the location and size of the tumor and its relationship with the prostate capsule (Fig. 4).

After discussion, a surgical treatment was conducted after fully explaining the treatment plan to the patient's family and obtaining their consent. After adequate preoperative preparation, the patient underwent laparoscopic radical prostatectomy on July 25, 2019. The prostate volume significantly increased during the operation, and the right seminal vesicle attachment mass had unclear boundaries. Surgical specimens were treated following the standard pathological procedures and analyzed by more than two experienced anatomical pathologists. Postoperative pathology revealed that the capsule of tumor tissue remained intact. The cut surface was pale, and no tumour was affected in the incised edge and bilateral seminal vesicle glands. Accordingly, a prostate leiomyosarcoma diagnosis (stage: T2cN0M0) was confirmed (Fig. 2B).

The patient was discharged four days after surgery. The patient could autonomously control urination after the catheter was removed on the seventh day after the operation. The pathological results suggested that the patient should receive adjuvant treatment, but he refused after a day. No tumor recurrence or metastasis was found after one year of follow-up with regular digital rectal examinations and abdominal MR scans.



FIG. 4. 3D reconstruction: Prostate was enlarged with abnormal shape, capsule of prostate was intact and no local invasion was observed.

### 3. Discussion

The rarity of adult prostate sarcoma makes clinical research difficult [10]. Therefore, knowledge of its clinical features, management, and prognosis has primarily been derived from case reports, small institutional series, and other genitourinary sites [11-13]. Prostate Leiomyosarcoma occurs in patients between 41 and 78 years (mean 61 years) [14, 15]. However, its pathogenesis is unknown. Kaufman et al. [16] defined prostate leiomyosarcoma as a localized or encapsulated space-occupying smooth muscle lesion with a diameter  $\geq$  1 cm containing different amounts of fibrous tissue no glandular components and occurs in or around the prostate. There are large differences in tumor size of prostate leiomyosarcoma, ranging between 3 cm to 21 cm [17]. Early symptoms of prostate leiomyosarcoma include frequent urination, dysuria, and urinary retention. However, they are often clinically ignored and not considered as prostate malignancy because they do not have typical clinical manifestations such as prostatic hard mass or elevated PSA. Therefore, the tumor is usually at an advanced stage during diagnosis, with poor differentiation and prognosis [18-21]. Its clinical diagnosis is complex mainly through pathological examination and immunohistochemical staining.

Cheville [22] conducted a clinical analysis of 23 prostate leiomyosarcoma cases and found that tumor cells express vimentin, SMA, and junction proteins, and up to 25% of tumor cells express cytokeratin. For the patient, imaging specialists determined prostate cancer PSA was normal and pathological examination showed heterotypic spindle cell. Immunohistochemical staining was positive for desmin, SMA, and vimentin, focally positive for CK, similar to the characteristics of the above case. The fibrosarcoma cells were also fusiform under a microscope, but the immunohistochemistry was mainly vimentin-positive and negative for desmin and SMA.

Presently, there is no clear treatment guideline for this disease. The current treatment methods include surgery, radiotherapy, and chemotherapy. According to domestic and foreign reports, surgical treatment combined with radiotherapy and chemotherapy is the main treatment [23]. For patients with clinically localized disease, complete surgical resection with negative margins is the major sarcoma treatment and has higher chances of finding the cure [24]. Some studies have suggested that multimodal therapy is more effective than surgical resection alone [17, 23]. However, its advantages are unknown because of the small sample sizes of the studies and variations in treatment. Surgical methods include radical prostatectomy, radical cystoprostatectomy, and total pelvic exenteration [20]. Common chemotherapy regimens are based on anthracyclines (doxorubicin or epirubicin) combined with alkylating agents (cyclophosphamide, ifosfamide, or dacarbazine) and/or vinca alkaloids (vinblastine or vincristine) [25]. Novel targeted agents such as imatinib, which is used to treat metastatic gastrointestinal stromal tumors, can also provide an alternative treatment approach for adult prostate sarcoma in the future [26]. Despite aggressive management, local control and survival have been poor [15].

The early symptoms of prostate leiomyosarcoma lack specificity and are easily overlooked or misdiagnosed as

benign prostatic hyperplasia. Leiomyosarcoma is usually a terminal illness during its diagnosis because of its rapid development. About a third of patients could have metastatic disease mainly in the lungs, liver, and bone [20, 21]. Overall survival is low, and one-year, three-year, and five-year survival rates are estimated to be 68%, 34%, and 26%, respectively [11]. Prostate leiomyosarcoma is one of the most severe prostate malignancies with a poor prognosis and a high risk of death. While the overall prognosis is poor, the prognosis is better in patients with no distant metastasis during the first presentation and in patients with localized diseases that are negative at the incisal edge under the microscope [20]. A good balance is required between the maximum amount of normal tissue preserved and the risk of positive surgical margins because of cancer focally extending beyond the prostate capsule increases the risk of positive surgical margins. The 3D reconstruction technology facilitates the development of detailed 3D virtual models of the prostate that identify the tumor and its relationship with the prostate capsule. Thus, intraoperative knowledge of the 3D location of cancer lesions can prevent the surgeon from conceptualizing the procedure when using two-dimensional (2D) preoperative images, potentially reducing this risk [27]. Thus, the application of 3D reconstruction technology is important in improving the prognosis of patients.

However, this case report has several limitations. First, the sample size was too small with only one case. Second, the follow-up time was short (one year) and the patient could not have fully recovered. Third, the patient only received surgical treatment and refused adjuvant therapy, thus the effect of adjuvant therapy could not be explained. However, this study has a few advantages. Before surgery, the location and size of the tumor can be determined to know if it breaks through the capsule using 3D reconstruction. The optimal resection range can also be determined using the 3D reconstruction during surgery to ensure negative resection margin, the maximum amount of normal tissue, and promote postoperative recovery and autonomous urination.

### 4. Conclusions

Prostate leiomyosarcoma is a malignant tumor with low incidence, rapid development, high malignancy, and poor prognosis. It has no standard treatment plan, and only comprehensive treatment based on surgery can be used. Preoperative three-dimensional reconstruction is used to determine tumor location, size, and invasion range, and is convenient for designing surgical plans and operations. In conclusion, early detection, diagnosis, and treatment and the application of 3D reconstruction technology have positive effects on prostate leiomyosarcoma prognosis.

## Abbreviations

2D, two-dimensional; 3D, Three-dimensional; CK, Cytokeratin; CT, computed tomography; MRI, magnetic resonance imaging; NSE, Neuron Specific Enolase; PSA, Prostate Specific Antigen; SMA, Smooth Muscle Actin; RMS, rhabdomyosarcoma; TAM, tumor associated material; Vim, Vimentin.

# Author contributions

- (I) Conception and design: Zhenli Gao
- (II) Administrative support: Zhenli Gao and Jitao Wu
- (III) Collection of materials of patients: Zhongbao Zhou
- (IV) Collection of literatures: Zhunan Xu and Tong Cai
- (V) Manuscript writing: Zhunan Xu and Tong Cai
- (VI) Final approval of manuscript: All authors

### Ethics approval and consent to participate

This is a case report and does not involve any experimental research, so I don't think it is necessary to apply for ethical permission.

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### **Conflict of interest**

The authors declare that there were no conflicts of interest.

### Consent

The case was reported with the patient's oral consent.

### References

- Siegel R, Naishadham D, Jemal A. Cancer statistics, 2013. CA: A Cancer Journal for Clinicians. 2013; 63: 11-30.
- [2] Rojas-Jiménez A, Otero-Garcia M, Mateos-Martin A. Stromal prostatic sarcoma: a rare tumor with rare clinical and imaging presentation. Journal of Radiology Case Reports. 2013; 7: 24-31.
- [3] Riley DS, Barber MS, Kienle GS, Aronson JK, von Schoen-Angerer T, Tugwell P, et al. CARE guidelines for case reports: explanation and elaboration document. Journal of Clinical Epidemiology. 2017; 89: 218-235.
- [4] Schubert C, van Langeveld MC, Donoso LA. Innovations in 3D printing: a 3D overview from optics to organs. British Journal of Ophthalmology. 2014; 98: 159-161.
- [5] Bird J. Exploring the 3D printing opportunity. The Financial Times. 2012.
- [6] George A. 3-D printed car is as strong as steel, half the weight, and nearing production. Wired. 2013. Available at: http: //www.wired.com/autopia/2013/02/3d-printed-car/ (Accessed: 27 February 2015).
- [7] Marks P. 3D printing: the world's first printed plane. New Scientist. 2011. Available at: http://www.newscientist.com/article/ dn20737-3d-printing-the-worlds-first-printedplane.html#Ux4SLx\_LI7x (Accessed: 01 August 2014).
- [8] Greenberg A. Meet the 'liberator': test-firing the world's first fully 3Dprinted gun. Forbes. 2013. Available at: http://www.forbes.com/ sites/andygreenberg/2013/05/05/meet-the-liberatortest-firing-the-worlds-first-fully-3d-printed-gun/ (Accessed: 14 June 2015).
- [9] Macguire E. Dutch architects build world's first 3D printed house. CNN. 2015. Available at: http://edition.cnn.com/2014/04/ 13/business/3d-printed-house-amsterdam/ (Accessed: 14 April 2015).



- [10] Mondaini N, Palli D, Saieva C, Nesi G, Franchi A, Ponchietti R, et al. Clinical characteristics and overall survival in genitourinary sarcomas treated with curative intent: a multicenter study. European Urology. 2005; 47: 468-473.
- [11] Humphrey PA. Angiosarcoma of the prostate. The Journal of Urology. 2012; 187: 684-685.
- [12] Ren FY, Lu JP, Wang J, Ye JJ, Shao CW, Wang MJ. Adult prostate sarcoma: radiological-clinical correlation. Clinical Radiology. 2009; 64: 171-177.
- [13] Boyle H, Fléchon A, Droz JP. Treatment of uncommon malignant tumours of the bladder. Current Opinion in Urology. 2011; 21: 309-314.
- [14] Zazzara M, Divenuto L, Scarcia M, Cardo G, Maselli FP, Ludovico GM. Leiomyosarcoma of prostate: case report and literature review. Urology Case Reports. 2017; 17: 4-6.
- [15] Vandoros GP, Manolidis T, Karamouzis MV, Gkermpesi M, Lambropoulou M, Papatsoris AG, *et al.* Leiomyosarcoma of the prostate: case report and review of 54 previously published cases. Sarcoma. 2008; 2008: 458709.
- [16] Kaufman JJ, Berneike RR. Leiomyoma of the prostate. The Journal of Urology. 1951; 65: 297-310.
- [17] Dotan ZA, Tal R, Golijanin D, Snyder ME, Antonescu C, Brennan MF, et al. Adult genitourinary sarcoma: the 25-year memorial Sloan-Kettering experience. Journal of Urology. 2006; 176: 2033-2039.
- [18] Janet NL, May A, Akins RS. Sarcoma of the prostate: a single institutional review. American Journal of Clinical Oncology. 2009; 32: 27-29.
- [19] Miedler JD, Maclennan GT. Leiomyosarcoma of the prostate. The

Journal of Urology. 2007; 178: 668.

- [20] Hossain D, Meiers I, Qian J, MacLennan GT, Bostwick DG. Prostatic leiomyoma with atypia: follow-up study of 10 cases. Annals of Diagnostic Pathology. 2008; 12: 328-332.
- [21] Sexton WJ, Lance RE, Reyes AO, Pisters PWT, Tu S, Pisters LL. Adult prostate sarcoma: the M. D. Anderson Cancer Center experience. Journal of Urology. 2001; 166: 521-525.
- [22] Cheville JC, Dundore PA, Nascimento AG, Meneses M, Kleer E, Farrow GM, et al. Leiomyosarcoma of the prostate. Report of 23 cases. Cancer. 1995; 76: 1422-1427.
- [23] Edge SB, Compton CC. The American Joint Committee on Cancer: the 7th edition of the AJCC cancer staging manual and the future of TNM. Annals of Surgical Oncology. 2010; 17: 1471-1474.
- [24] Clark MA, Fisher C, Judson I, Thomas JM. Soft-tissue sarcomas in adults. The New England Journal of Medicine. 2005; 353: 701-711.
- [25] Lin M, Hsiao H. Promising effects of pazopanib with radiation on an advanced prostate leiomyosarcoma after failure of systemic chemotherapy. The Kaohsiung Journal of Medical Sciences. 2019; 35: 317-318.
- [26] Heinrich MC, Corless CL, Demetri GD, Blanke CD, von Mehren M, Joensuu H, et al. Kinase mutations and imatinib response in patients with metastatic gastrointestinal stromal tumor. Journal of Clinical Oncology. 2003; 21: 4342-4349.
- [27] Porpiglia F, Bertolo R, Checcucci E, Amparore D, Autorino R, Dasgupta P, *et al.* Development and validation of 3D printed virtual models for robot-assisted radical prostatectomy and partial nephrectomy: urologists' and patients' perception. World Journal of Urology. 2018; 36: 201-207.