International Journal of Research in Medical Sciences
Shah A et al. Int J Res Med Sci. 2024 Jan;12(1):280-283
www.msjonline.org
pISSN 2320-6071 | eISSN 2320-6012

DOI: https://dx.doi.org/10.18203/2320-6012.ijrms20234020

Case Report

When pathology defies expectations: a surprising diagnosis of prostatic leiomyosarcoma

Abhijit Shah, Anshuman Singh, Kasi V. Gali, Vijay Gunashekar, Padmaraj Hegde*

Department of Urology and Renal Transplant, Kasturba Medical College, Manipal, Karnataka, India

Received: 07 November 2023
Accepted: 07 December 2023

***Correspondence:**
Dr. Padmaraj Hegde,
E-mail: padmaraj.hegde@manipal.edu

ABSTRACT

Prostatic leiomyosarcoma is an exceedingly rare malignant tumour arising from smooth muscle cells in the prostate gland. It ranks quite low down on the possible differentials of bladder outlet obstruction. We present a case of a 38-year-old male who initially presented with acute urinary retention and was subsequently diagnosed with prostatic leiomyosarcoma. Following histopathological confirmation, metastatic work-up was performed and patient planned for radiation and surgical cure. Patient on follow-up is well and has resumed normal daily work. This case report aims to increase awareness of this rare malignancy, discuss the diagnostic challenges faced with a highly common clinical spectrum at the onset, present the treatment strategies employed, and discuss the patient's clinical outcomes. We emphasise the importance of reporting rare cases like this to contribute to the existing literature and enhance the understanding of prostatic leiomyosarcoma.

Keywords: Prostatic neoplasm, Sarcoma, Leomyosarcoma, Lower urinary tract symptoms

INTRODUCTION

Prostatic leiomyosarcoma is an uncommon malignancy originating from smooth muscle cells within the prostate gland.[1] It represents a diagnostic challenge due to its rarity and nonspecific clinical presentation.[1] We present a case of prostatic leiomyosarcoma in a 38-year-old male who initially presented with acute urinary retention. This case report aims to contribute to the existing medical literature by highlighting the diagnostic challenges encountered, the treatment strategies employed, and the patient's clinical outcomes. The rarity of prostatic leiomyosarcoma and the heterogeneity in its clinical presentation underscore the importance of reporting such cases to enhance awareness and understanding of this malignancy.

CASE REPORT

A 38-year-old male presented to our outpatient department with complaints of acute urinary retention for which he required catheterization. The patient reported a history of poor urinary stream and nocturia for the past three months, along with recent onset dysuria and intermittent hematuria. On examination, systemic findings were unremarkable, and a digital rectal exam revealed a grade 1 prostate with mobile mucosa and firm margins.

Routine investigations, including urine examination, were essentially normal. Ultrasound revealed a prostate size of 40cc with normal upper tracts and no evidence of clots or debris in the bladder. The prostate-specific antigen (PSA) level was 2 ng/ml.

As the patient failed a trial without catheter and continued to experience persistent symptoms, he underwent transurethral resection of the prostate (TURP). Histopathological examination of the resected tissue revealed the presence of spindle cells within an ill-circumscribed lesion with a few mitotic figures (1-2/10 high-power fields). Immunohistochemistry confirmed the diagnosis of prostatic leiomyosarcoma, with tumour cells diffusely positive for smooth muscle actin (SMA) and negative for cytokeratin (CK) (Figure 1).

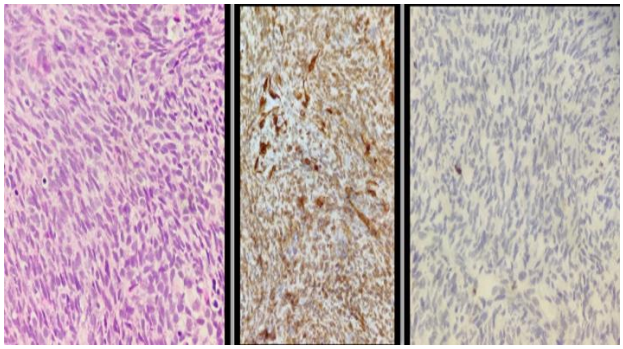

Figure 1: Left to right- H&E, SMA- 400x, CK-400x.

An MRI of the pelvis was performed followed by a whole body F18 FDG-PET/CT (Figure 2). Magnetic resonance imaging (MRI) revealed diffuse restriction and early contrast enhancement in the posterior part of the prostate, extending into the rectum and infiltrating the seminal vesicles. F18 FDG-PET/CT showed increased uptake in the mesorectal and iliac nodes, indicating metastatic involvement (Figure 3). Following these findings, due consent was taken from patient for possible reproduction of this case as public academic text and written assurance of maintaining anonymity was given to the patient.

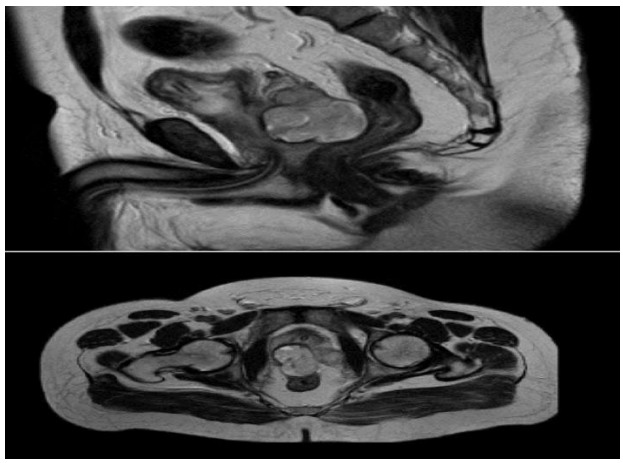

Figure 2: MRI of the pelvis.

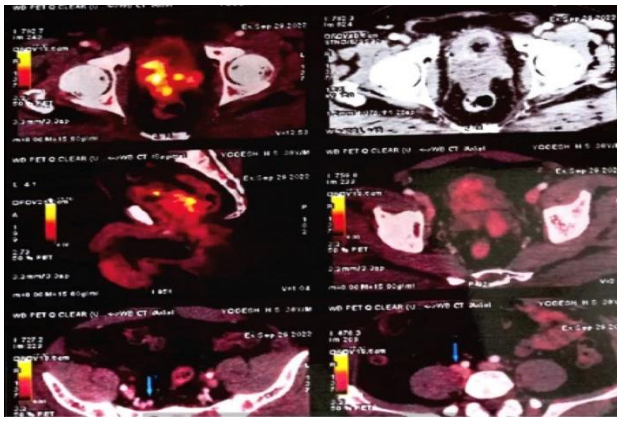

Figure 3: F18 FDG-PET/CT showed increased uptake in the mesorectal and iliac nodes.

A multidisciplinary tumour board convened to discuss the case and as per their recommendation, the patient was scheduled to undergo neoadjuvant external beam radiotherapy (EBRT) at a dose of 50 Gy delivered in 25 daily fractions, followed by definitive surgery. However, for logistical reasons, the patient opted to receive radiation therapy at a centre closer to their home. Following the completion of the 25 fractions of EBRT, the patient underwent open radical prostatectomy at a different centre. On follow-up at our OPD, at 5th month the patient was doing well with infrequent complaints of incontinence that has symptomatically improved over the past few months and he has resumed his office work, with a planned PET scan the following month.

DISCUSSION

Prostatic leiomyosarcoma is an exceedingly rare malignant tumour arising from smooth muscle cells within the prostate gland. Despite its rarity, there have been several case reports and literature reviews published on this topic, shedding light on the clinical, diagnostic, and therapeutic aspects of this aggressive neoplasm. In this discussion, we compare our presented case report with the previously published literature, emphasizing the unique features and treatment strategies employed.

Comparing our case with the literature, it is consistent with the findings reported in the literature regarding the clinical presentation of prostatic leiomyosarcoma.[4,5] It becomes apparent that prostatic leiomyosarcoma presents a diagnostic challenge due to its nonspecific clinical presentation. Our patient initially experienced lower urinary tract symptoms, including poor urinary stream, nocturia, dysuria, and intermittent hematuria. Similar symptoms have been reported in the literature by Vandoros et al and Singh et al highlighting the diagnostic difficulties due to non-specific presenting complaints.[1,2]

Differential diagnoses of prostatic leiomyosarcoma include nodular hyperplasia of the prostate with atypical changes, postoperative spindle cell nodules, rectal gastrointestinal stromal tumor (GIST), stromal sarcoma of the prostate, sarcomatoid carcinoma of the prostate, and leiomyomas.[3] Immunohistochemical staining plays a crucial role in distinguishing these conditions. Nodular hyperplasia of the prostate typically exhibits no invasion, mitotic figures, and shows positive staining for CD10.[4] Postoperative spindle cell nodules are characterized by cellular tissue with high mitotic activity, extravasation of red blood cells, and relatively small size.[1,5] Rectal GIST shows positive staining for CD117, CD34, vimentin, and may exhibit skeinoid fibers.[11] Stromal sarcoma of the prostate is positive for CD56, synaptophysin, and CD99.[6,7]

Sarcomatoid carcinoma of the prostate presents as a biphasic tumor with positive staining for cytokeratin and prostate-specific antigen (PSA) in the epithelial component. Leiomyomas demonstrate well-organized fascicles of spindle cells and positive staining for desmin,

actin, and androgen receptor.[8,9] In our case, the diagnosis was confirmed through histopathological examination and immunohistochemistry of the specimen obtained after transurethral resection of the prostate. The immunohistochemical analysis demonstrated diffuse positivity for smooth muscle actin (SMA) and negativity for cytokeratin (CK) which help to differentiate this diagnosis from other similar lesions.[1]

The optimal treatment strategies for prostatic leiomyosarcoma remain controversial due to limited evidence and the aggressive nature of the disease.[1] Surgical resection is considered the preferred approach in cases where the disease is localized and has well-defined margins.[1,10] However, external beam radiotherapy (EBRT) plays a significant role in patients who are not suitable for surgery or when the tumour needs to be downsized prior to surgical removal.[1,11] Therefore, a multimodality treatment regimen with a multidisciplinary approach is strongly recommended.[1,12] Poonia et al have attempted to develop a management algorithm for prostatic leiomyosarcoma, proposing upfront surgery with or without adjuvant chemotherapy or radiotherapy for resectable disease.[12] In our case, the patient initially underwent transurethral resection of the prostate (TURP) after failing a trial without a catheter. However, disease progression was observed during follow-up, as evidenced by recurrent acute urinary retention and metastatic involvement detected on F18 FDG-PET/CT.

Subsequently, the multidisciplinary tumour board recommended neoadjuvant EBRT followed by definitive surgery for the patient, aligning with the treatment approach described by Poonia et al in their case report, which emphasized the necessity for individualized and multidisciplinary management strategies.[12]

Given the rarity of prostatic leiomyosarcoma and the limited number of cases reported in the literature, evidence-based treatment guidelines are lacking. As a result, treatment decisions should be made on a case-by-case basis, considering the patient's overall health status, disease stage, tumour characteristics, and the expertise of the treating physicians. Close collaboration between urologists and radiation oncologists is essential for optimizing treatment outcomes for patients with prostatic leiomyosarcoma.

The presented case contributes to the existing literature by providing insights into the clinical course and treatment outcomes of prostatic leiomyosarcoma. However, it is important to acknowledge the limitations of this report, which include a single-patient focus, and the absence of long-term follow-up data. Another limitation exists in our inability to perform a cross-sectional imaging at the onset for haematuria due to unwillingness of patient which may have altered the management at the start itself. Further research and collaboration are needed to accumulate more cases and establish evidence-based guidelines for the management of this rare malignancy.

CONCLUSION

In conclusion, prostatic leiomyosarcoma is a rare malignancy with diagnostic challenges and limited treatment guidelines. Our case report adds to the existing literature, presenting a unique patient profile and treatment approach. Collaborative efforts and continued reporting of such cases will enhance understanding, improve diagnostic accuracy, and guide optimal management strategies for patients with this diagnosis.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

REFERENCES

1. Cheville JC, Dundore PA, Nascimento AG, Meneses M, Kleer E, Farrow G, et al. Leiomyosarcoma of the Prostate Report of 23 Cases. Cancer. 1995;76(8):1422-7.
2. Zazzara M, Divenuto L, Scarcia M, Cardo G, Maselli FP, Ludovico GM. Leiomyosarcoma of prostate: Case report and literature review. Urol Case Rep. 2018;17:4.
3. Venyo AKG. A Review of the Literature on Primary Leiomyosarcoma of the Prostate Gland. Adv Urol. 2015;485786.
4. Tawfic S, Niehans GA, Manivel JC. The pattern of CD10 expression in selected pathologic entities of the prostate gland. Hum Pathol. 2003;34(5):450-6.
5. Pathology Outlines. Postoperative spindle cell nodule. 2014. Available at: https://www.pathologyoutlines.com/topic/bladderpostopspindle.html. Accessed on 12 September 2023.
6. Wick MR, Young RH, Malvesta R, Beebe DS, Hansen JJ, Dehner LP. Prostatic carcinosarcomas. Clinical, histologic, and immunohistochemical data on two cases, with a review of the literature. Am J Clin Pathol. 1989;92(2):131-9.
7. Yamazaki H, Ohyama T, Tsuboi T, Taoka Y, Kohguchi D, Iguchi H, et al. Prostatic stromal sarcoma with neuroectodermal differentiation. Diagn Pathol. 2012;7(1):173.
8. Hossain D, Meiers I, Qian J, MacLennan GT, Bostwick DG. Prostatic leiomyoma with atypia: follow-up study of 10 cases. Ann Diagn Pathol. 2008;12(5):328-32.
9. Hansel DE, Herawi M, Montgomery E, Epstein JI. Spindle cell lesions of the adult prostate. Modern Pathol. 2006;20(1):148-58.
10. Xu ZN, Cai T, Zhou ZB, Wu JT, Gao ZL. Prostate leiomyosarcoma treatment using three-dimensional reconstruction: a case report and literature review. J Mens Health. 2021;17(2):148-53.
11. 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;458709.

12. Poonia DDR, Singh DA, Sunil H, Gandhi DJ, Rawal DS. Management of Leiomyosarcoma of the Prostate: Unraveling a Hoax. Curr Prob Cancer. 2021;3:100068.

Cite this article as: Shah A, Singh A, Gali KV, Gunashekhar V, Hegde P. When pathology defies expectations: a surprising diagnosis of prostatic leiomyosarcoma. Int J Res Med Sci 2024;12:280-3.