

Recurrent Hematuria, Urinary Tract Infections, and Urinary Retention: A Rare Presentation of Prostate Leiomyosarcoma

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Abstract:

Prostatic leiomyosarcoma is quite rare. There is little consensus on the best treatment for these aggressive tumors. They are most often characterized by obstructive lower urinary tract symptoms without an elevated serum prostate-specific antigen level. A high degree of skepticism may help in detecting them in a timely manner since they lack specific clinical or imaging features. It can be diagnosed by immunohistochemical staining as, seen in our patient. Despite the lack of definitive treatment options, innovative multimodal treatment strategies may significantly improve the prognosis of patients suffering from this tumor. This case report describes a leiomyosarcoma of the prostate with metastasis.

Introduction:

Prostate leiomyosarcoma is reported to be accounted for 0.1% of all prostate malignancies. It is a rare, aggressive tumor with fewer than 200 cases reported [1]; hence, treatment guidelines are not well established. It was accompanied by non-specific symptoms similar to those associated with benign prostatic hyperplasia. Our patient reported having recurrent episodes of hematuria. We present a rare case of prostate leiomyosarcoma treated with chemotherapy and a literature review that discusses clinical features, diagnostic methods, and therapeutic options.

Case presentation

A 61-year-old male with no significant past medical history presented to the emergency department with suprapubic discomfort and urinary retention. The patient has a twenty-five-pack-year smoking history, a family history of breast, pancreatic, and lung cancer, and no family history of prostate cancer. A pelvic/bladder ultrasound revealed an enlarged prostate gland measuring 8.6 cm x 7.9 cm x 9 cm, which was heterogenous with a weight of 320 g. PSA was measured to be 1.4 ng/mL. The patient continued to have frequent visits to the emergency department for lower urinary tract symptoms, after which he underwent an elective simple open prostatectomy. There was a 2.5 cm circumferential necrotic appearance to the right hemiprostate and asymmetric growth on the right side. In addition, pathology reported an incidental finding of leiomyosarcoma.

Immunohistochemical stains showed calponin (+ve), SMA (focal), Desmin (patchy), smooth muscle myosin heavy chain (+ve), caldesmon (+ve,) vimentin patchy, CD34 (-ve) and PR (-ve), CK5/6 (-ve). In addition, TSC2 and BRCA 1 genomic alternations were detected. The post-surgical course was complicated by hematuria, which was resolved with continuous bladder irrigation.

Computed tomography (CT) of the chest/abdomen and pelvis (CTAP) showed multiple nodular densities in the left and right lungs. A lobular contour cyst in the liver measures about 1 cm x 0.59 cm, along with cysts in the right and left kidneys. In addition, tiny nodes in the retroperitoneum with prominent vessels are noted. There was a small amount of intra-abdominal and pelvic ascites noted. The prostate gland, enlarged with low attenuated lesions in the transitional and central zones, measures about 4.6 cm x 2.9 cm on the right, and the left measures about 1.9 cm in the transitional zone. The prostate measures about 8.1 cm x 8.3 cm x 7.1 cm. NM bone scan revealed heterogeneous uptake in the right posterior iliac spine, right scapular region, and right distal region, indicating osseous metastases. The patient was started on chemotherapy with a doxorubicin regimen every three weeks.

The patient continued to have recurrent hospitalizations for urinary retention, suprapubic pain, worsening hematuria, and urinary tract infections. He was treated with antibiotics and multiple blood transfusions. Further course was complicated by left-sided hydronephrosis, requiring left-sided nephrostomy tube placement.

The patient is currently being managed with an outpatient chemotherapy regimen.

Discussion

This case study highlights the distinct challenges clinicians face when diagnosing and managing prostatic leiomyosarcomas and determining the etiology of concurrent urologic symptoms over the course of its management. Prostate leiomyosarcomas are extremely rare, as they compose 0.1% of primary prostate tumors; sarcomas account for 1% of malignancies, with 5% of sarcomas arising from the genitourinary tract [2]. In each of the rare cases, the condition may present with a plethora of non-specific genitourinary symptoms such as recurrent hematuria, urinary frequency or urgency, recurrent urinary tract infections (UTIs), urinary retention, or generalized suprapubic pain. The FNCLCC grading system and histopathological examination

are typically used for the definitive diagnosis. The tumor histology is notable for marked necrosis, hypercellularity, nuclear pleomorphism without degenerative features, positive histochemical stains such as smooth muscle actin, Desmin, vimentin, calponin, and CD34, and is commonly differentiated from other prostatic stromal lesions known as STUMP [3]. In contrast to more common tumors of the prostate, normal or slightly elevated PSA values can be noted in prostate leiomyosarcomas, as the tumor has a nonepithelial origin. Although no standard recommendations are established to treat prostate leiomyosarcoma, a wide variety of treatments may be used and can include surgery, if operable, as well as chemotherapy and radiotherapy. However, the long-term survival rate and overall prognosis remains poor [4].

The described case features an unusual presentation of a rare prostate malignancy associated with multiple postoperative hospitalizations for recurrent hematuria, urinary retention, suprapubic pain, and urinary tract infections (UTIs). Although urinary frequency and urgency are commonly reported symptoms in patients with prostate leiomyosarcomas, recurrent hematuria and urinary retention have only been reported in a handful of isolated cases. In this case of a 61-year-old male with a strong smoking history and a normal PSA level, hematuria may be a rare obvious symptom that can delay its diagnosis [5]. This patient's unique symptoms of recurrent hematuria and acute urinary retention may be associated with either the malignancy or due to a multitude of other factors.

The urinary retention and recurrent UTIs may have occurred secondary to a significantly enlarged prostate and underlying BPH. In contrast, the recurrent hematuria that began shortly after the prostatectomy may be attributed to a documented adverse event from either the operation [6], post-operational radiotherapy [7], or doxorubicin treatments [8]. Despite the multifactorial etiology of the recurrent hematuria, the hematuria was most likely caused directly by the leiomyosarcoma. This is because recurrent hematuria has been linked to prostate leiomyosarcomas due to hypervascularity [9, 10]. The involvement of the anterior transitional zone and right distal ureter in this patient's tumor, rather than the commonly affected peripheral zone in other prostate malignancies, may have also contributed to the resulting hematuria. In addition, dysuria and stranguria, or symptoms of bladder outlet obstruction, have been noted to occur with prostate sarcomas [2, 4].

As prostate leiomyosarcomas with recurrent episodes of hematuria are rare, it is of note that this case may have an etiological genetic component. While the patient denied any family history of prostate cancer or bleeding disorders, the patient's sister and maternal grandmother had breast cancer in their early 60s and 80s, respectively. In addition, his mother was diagnosed with pancreatic cancer at 75. This may be due to a germline mutation through his maternal lineage, as prostate cancers such as leiomyosarcomas have been associated with a family history of breast and pancreatic cancers diagnosed at a younger age [11].

Prostate leiomyosarcomas can have several systemic manifestations and may be difficult to diagnose in certain situations. Aside from the clinical symptoms due to the primary prostatic growth, the metastatic and extra-prostatic manifestations of the sarcoma, such as the ascites and osteoblastic pulmonary lesions, can be debilitating and have a significant impact on the quality-of-life of the affected patient. For this reason, its complex pathology requires clinicians to be aware of the masquerading effects of other genitourinary conditions and postoperative adverse events on potential neoplastic symptoms and the multifactorial etiology of non-specific symptoms such as hematuria and urinary retention.

The constellation of typical constitutional symptoms and physical exam findings, including recurrent hematuria, urinary retention, suprapubic pain, and UTIs, is non-specific and may be due to either infectious, iatrogenic, or neoplastic conditions. For this reason, determining the etiology of these symptoms is paramount in establishing a timely diagnosis and preventing significant delays in disease management and treatment. In addition, diagnostic tests such as a timely abdominal CT scan can detect malignant ascites that may significantly affect the patient's prognosis and management. Although prostate leiomyosarcomas are not often seen in clinical practice, it is crucial to be aware of its presentation as an accurate diagnosis may improve the patient's quality and life expectancy.

Conclusion:

Detecting prostate leiomyosarcoma can be difficult due to the variability in clinical presentations, as well as the lack of clear clinical signs of malignancy. Leiomyosarcoma of the prostate is a rare neoplasm, often presents in the metastatic stage with normal PSA levels and usually takes an aggressive course. Therefore, a multidisciplinary approach is essential for the appropriate management of this cancer.

Authorship:

Sowmya Sagireddy searched the literature, wrote, and revised the manuscript. Chiya Abramowitz wrote discussion and searched the literature. Sai Sagireddy and Marc Sukhoo-Pertab revised and edited the manuscript. Mark Sonnenschine approved the final version and is the article's guarantors. All authors certify that they contributed sufficiently to the intellectual content and data analysis. Each author has reviewed the final version of the manuscript and approves it for publication.

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Not applicable

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This study protocol was reviewed and the need for approval was waived by the ethics committee at South Brooklyn Health.

Consent for publication:

Written informed consent was obtained from the patient for publication of this case report.

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