CASE REPORT

A rare case of urinary bladder leiomyosarcoma accompanied by prostate cancer

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Non-urothelial neoplasms of the bladder account for less than 5% of all bladder tumors. Sarcoma constitutes the most usual mesenchymal malignancy of the bladder, with leiomyosarcomas being the most common type of sarcoma in adults. A coexistence of this rare tumor with another malignancy has not been reported, to our knowledge. This report demonstrates a case of high-grade bladder leiomyosarcoma cancer with prostate cancer in a 72-year-old patient. The most striking features of this case include the rapid disease progression that confirms the highly aggressive nature of this uncommon disease.

Key Words: bladder leiomyosarcoma, prostate cancer, prognosis

Introduction

Prostate cancer is the fourth most common cancer in men, with incidence and mortality rates that vary markedly among and within different countries. Since

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Address correspondence to Dr. Apostolos P. Labanaris, Department of Urology, Martha Maria Medical Center, Stadenstrasse 58, 90409 Nürnberg, Germany the early 1990s, screening tests and improved treatments have been associated with dramatic shifts in the incidence, stage at diagnosis, and mortality of this disease. On the other hand, leiomyosarcomas of the bladder are rare tumors and have always been considered as a highly aggressive entities portending a grim prognosis.^{1,2} Little is known about their origin, clinicopathologic presentation and the survival factors associated with them. We present, to our knowledge, the first reported case of bladder leiomyosarcoma accompanied by prostate cancer.

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Case report

In November 2006, a 72-year-old man presented to our department with intermittent gross hematuria accompanied by symptoms of vesical irritability. The patient had not undergone radiotherapy or chemotherapy for previous disease and his history revealed no smoking habits. Urinalysis revealed numerous red blood cells, and the urine culture was negative. His serum prostate-specific antigen level was slightly elevated (4.750 ng/ml) but no abnormalities were found on digital rectal examination, and his history of first-degree relatives with prostate cancer was negative. Abdominal ultrasonography revealed a solid lesion, approximately 5 cm in length, located on the base of the bladder close to the left urethral orifice, without associated ureterohydronephrosis. A staging computed tomography scan of the abdomen and pelvis, a bone scan, chest radiography, and magnetic resonance imaging did not provide information leading to a diagnosis, Figure 1. Cystoscopy, with transurethral resection of the bladder tumor, was initially performed, and leiomyosarcoma was initially diagnosed on the basis of examination of the transurethral specimen. Additionally, transrectal ultrasound-guided needle biopsy was performed, which revealed a T1c G2 Gleason 6 (3+3) prostatic carcinoma.

After 2 weeks, the patient underwent a definitive surgical procedure. Radical cystoprostatectomy coupled with pelvic lymphadenectomy and orthotopic urinary diversion (Mainz II neobladder) was performed. The final pathology results from the cystoprostatectomy specimen exhibited a high-grade bladder leiomyosarcoma stage 3, Figure 2. The pathologic stage was assigned using the Memorial



Figure 1. MRI of the patient's bladder leiomyosarcoma.

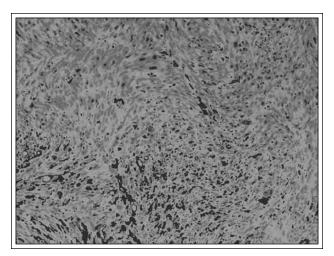


Figure 2. Microscopic appearance of the patient's bladder leiomyosarcoma.

Sloan-Kettering Cancer Center grading system;³ the urinary bladder leiomyosarcoma was accompanied by a T2b G2 N0 R0 M0 Gleason 6 (3+3) prostatic carcinoma and positive lymph nodes (2/39). The positive lymph nodes were the result of the patient's bladder leiomyosarcoma and not his prostate cancer, since they exhibited infiltration of a mesenchymal malignancy. Complete resection with negative surgical margins were achieved for both prostate and bladder specimens.

The adjuvant radiochemotherapeutic regimen that was scheduled for the patient was doxorubicin and ifosfamide combined with radiotherapy, but the patient refused additional therapy. The patient passed away 9 months later, after lung and liver metastases were diagnosed.

Discussion

To our knowledge, this is the first report of a patient with a bladder leiomyosarcoma accompanied by prostate cancer. Although prostate cancer is the fourth most common cancer in men, non-urothelial neoplasms of the bladder account for less than 5% of all bladder tumors.⁴ Strong evidence supports the fact that the incidence of leiomyosarcomas increases in patients receiving local pelvic radiotherapy or systemic chemotherapy treatment for different neoplasms.^{5,6} Contrary to patients with transitional cell carcinoma, tobacco use does not seem to increase the incidence of bladder leiomyosarcomas.⁷

Prostate cancer is known to be associated with genetic factors. Indeed, a number of case-control studies have demonstrated familial clustering of prostate cancer, suggesting that first-degree relatives of men with prostate cancer have a significantly higher risk of developing prostate cancer.⁸

Although surgical treatment of prostate cancer offers excellent local control and a reasonable expectation of cure — of course, always depending on clinical stage, Gleason grade, serum prostate-specific antigen level and positive surgical margins — leiomyosarcoma of the bladder portends a grim prognosis. Mackenzie et al in 1968, reported on 11 patients who survived longer than 3 years after diagnosis. 9 Nowadays, contemporary studies suggest that these tumors may have a better prognosis than once believed, with a remarkable 5-year disease specific survival rate of 62%. 10

The current patient's demographic characteristics and clinical presentation were similar to those in previous reports of patients with both bladder leiomyosarcomas and prostate cancer. He was in his seventh decade when he presented with hematuria and vesical irritability, and his clinical stage at presentation was T3. Additionally, when a transrectal ultrasound-guided needle biopsy was performed, it revealed a T1c G2 Gleason 6 prostatic carcinoma. No obstructive uropathy due to the size of his primary tumor was noted. This relatively uncommon tumor tends to invade locally, causing obstruction of the ureteral orifices. In most patients, when detected, the tumors exhibit advanced pathologic stage, with less than 30% having a stage T1 tumor.¹¹

Leiomyosarcomas require aggressive surgical extirpation, and when surgical resection is possible, radical cystectomy with wide margins is the "golden rule" and should be performed. Strict adherence to standard surgical technique (radical excision of the tumor with en bloc removal of the bladder to encompass the prostate and seminal vesicles in men, and the uterus, cervix, and vaginal cuff in women) have resulted in low rates of positive surgical margins and low rates of local tumor recurrence. Transurethral resection of the tumor, in combination with radiotherapy and chemotherapy, could be performed in individual patients (small tumors and/or inoperable patients), but does not offer better longterm results than surgery. Partial cystectomy could also be considered when the tumor is < 5 cm in diameter and when the surgeon is able to obtain cancer-free surgical margins, but it does not offer satisfying long-term results.12

Lymphovascular invasion and lymph node metastases are associated with a low disease-specific survival, and distant metastases overall recurrence rate is estimated to occur in about 53% of patients,

with the most common sites of metastases being lungs, liver, bone, and brain.⁷ Indeed, this patient passed away after only 9 months and after lung and liver metastasis were diagnosed. Of course, he had declined adjuvant radio-chemotherapy, but a 5-year disease specific survival rate in patients with positive lymph nodes is very disappointing.

Radical prostatectomy in patients with clinically localized prostate cancer is the "golden rule." Recent studies suggest that patients with T1–T2 NX MO prostate cancer who undergo radical prostatectomy display 5- and 10-year non-progression rates of 78% and 75%, respectively.¹³

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