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Case Report

Priapism as The Initial Presentation of Prostate Cancer Recurrence: A Case Report and Review of the Literature

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Abstract:

Introduction: Prostate cancer is one of the two most common non-cutaneous cancers in men. Its presentation might have unusual symptoms, which could have caused the wrong initial diagnosis.

Case Presentation: This study presents a case of prostate cancer complaining of persistent pain and erection of the penis. He had been treated with radical prostatectomy five years before his presentation. The patient had not followed his treatment for the cancer for the last four years. The patient was admitted to the hospital, and urgent imaging of the pelvis with magnetic resonance imaging showed extensive involvement of lymph nodes and corpus cavernosa. Management started with the combination of anti-androgenic and radiotherapy, which was effective and caused significant clinical improvement in the patient's condition and improved laboratory and radiologic markers.

Discussion: Prostate cancer recurrence can present with unusual symptoms considering its slow progression and involvement of the various structures. Malignant priapism is the involvement of the cavernosal system by a solid tumor. This is an indicator of a poor prognosis, and most patients' life expectancies are shorter than one year.

Clinical Key Message

Priapism can have a wide range of etiologies, and it can also be an indicator of malignancy progression through the genitourinary systems. In patients with a history of prostate cancer or those with a risk of this malignancy presented by priapism, appropriate diagnostic evaluation regarding this cancer should be performed. More importantly, patients treated for prostate cancer should be followed up regularly, and it should be mentioned that this cancer can slowly recur without any symptoms until it reaches a high stage.

Keywords: prostate cancer; metastasis; malignant priapism; radiotherapy; anti-androgen; case report

Introduction

Prostate cancer is one of the two most common non-cutaneous cancers in men and the fifth main cause of death among both genders in the world (1). Prostate cancer can recur after several years of treatment, and while it has been implied that the malignancy is cured. In many patients, the first symptoms of cancer emergence can emerge≥5 years after the radical prostatectomy (2). The other feature of prostate cancer is being asymptomatic for a long time due to its slow progression and the capability to remain dormant for several years before causing any detectable symptom or any sign in the patient's physical exam (3). The main location for the remaining cancerous cells, which cause the recurrence, is the prostate bed and its surrounding structures, and it

usually involves the surrounding lymph nodes, bones, lungs, and liver as the main locations of the initial metastasis (4). However, the recurrence does not follow a predictable pattern and can present with unusual and rare manifestations (5).

Priapism is prolonged penis erection which has occurred without any sexual stimulation and is unresponsive to conventional methods of treatment for more than four hours. This condition is classified to the three groups of high-flow (non-ischemic), low-flow (ischemic), and stuttering priapism (6). However, malignant priapism is a different entity which its mechanism has yet to be understood completely but the most probable scenario suggested for that is the invasion of solid tumors to the

cavernosal system (7). The spreading route in which cancerous cells spread to penis can be lymphatic or hematogenous leading to arterial or veno-occlusive conditions. Malignant priapism can be either ischemic or non-ischemic (8). Only about one-fourth of patients with malignant involvement of the penis present with priapism and they are mostly caused by genitourinary cancers specifically, bladder, prostate and kidney cancers (9). The presentation of the prostate cancer relapse with priapism is rarely seen and it is an indicator of end-stage and poor prognosis (10). Their life-expectancy is usually less than one-year (8).

2. Case Presentation

A 64-year-old man presented to his physician complaining of intermittent firm and painful penis began three months before his presentation. He had tried putting cold water, and ice to treat the condition which had not worked. The pain episodes had continued for different periods of time and was not related to sexual trigger. He was treated for a prostate cancer five years before presentation with radical prostatectomy. He had also followed-up the treatment for one-year after the surgery and since there were no rise in his prostate specific marker (PSA) or recurrence of any symptoms he had decided to discontinue regular follow up visits. The patient also had remarkable medical histories including ischemic heart disease, taking aspirin 81 mg/daily, and atorvastatin 40 mg every night. He had been smoker (roughly one packet per day) for more than 40 years. Both of his parents had the history of hypertension and chronic coronary disease and the patient's father had died due to metastatic colon cancer when he was 75 years old. Five years ago, Transrectal ultrasound showed an enlarged prostate; he noticed the cancer by screening by the PSA which was 4.5. A transurethral resection of the prostate (TURP) was performed revealing Gleason 4 + 3=7 adenocarcinomas of the prostate. After the surgery the PSA had declined to the undetectable level and remained without any rise until one year after the prostatectomy. He had no followup visit and no abnormal symptom suggesting recurrence during the last 4 years except a weight loss of 10 kilograms (declining from 85 to 75kg) during the last year. He attributed the wright loss to regular daily walking.

In his physical exam erection and swelling was evident and the digital rectal exam showed several irregular firm masses. No other remarkable finding was detected on his physical exam.

Considering the patient's condition, he was evaluated thoroughly starting by testing full blood tests, including PSA, and urgent doppler ultrasonography of the penis, which showed the low flow due to the blockage of corpus cavernosa by malignant cells. An urgent magnetic resonance imaging (MRI) of the pelvis showed a large irregular mass in the prostate bed despite the prostatectomy being performed five years ago, and expansion of the cancerous cells to multiple pelvic lymph nodes, pelvic bone, and corpus cavernosa. The PSA level become back to be 48.5ng/ml. Regarding the high risk of metastasis, a bone scan was performed which showed several sclerotic and lytic bone lesions in pelvic bone and lumbar spine. The diagnosis and related details were discussed with the patient, and the therapeutic options were discussed. However, he did not consent for any resection and surgical management and asked for medical treatment. Leuprolide injection was initiated the same day along with an oral intake of bicalutamide 150 mg/day. The patient was also advised to initiate pelvic radiotherapy as soon as possible.

The follow-up visit three months after the initiation of therapy showed remarkable improvement. The patient had a single injection of Diphereline (triptorelin embonate, 22.5mg) along with the daily bicalutamide tablets and 40 sessions of pelvic radiotherapy which was completed during the eight weeks. The symptoms have been regressed and the episodes were repeating with lower frequency and for a shorter period of time. PSA level had been declined to 9.8ng/ml and no abnormal laboratory data was detected in the full evaluation of blood markers, kidney and liver enzymes and urine sample.

3. Discussion

Magnant priapism is one of the rarely reported conditions and usually indicates the advanced stage cancer, poor prognosis, and short life expectancy. A study with 400 participants concluded that the average lifeexpectancy of malignant priapism was 9 months (11). However, the prognosis largely depends on the source of malignancy and in some cases, there are more available options for the management of the condition (5). Radiotherapy, chemotherapy, and penectomy are usually offered strategies, while the penectomy is mostly kept as the last option (12). In prostate cancer another option is hormone deprivation therapies which cut the source of cancerous cells growing stimulator such as antiandrogens, gonadotropin-releasing hormone (GnRH) agonists and 5α-reductase inhibitors, however their efficacy are not approved yet (13). Although pelvic radiotherapy have shown promising outcome, it can be accompanied by anatomical changes which might be troublesome for voiding of the patients (14). Hormonal therapies, on the other hand, have shown that they can affect the prognosis of prostate cancer and prolong the life expectancy (15). However, some studies claim that they cannot affect the local symptoms significantly (10). However, our patient responded profoundly to androgen deprivation therapy and the symptoms regressed remarkably after the initiation of the medications.

Xing et al. reported a case of prostate cancer metastasized to penis and caused priapism which was treated with a high-dose palliative radiotherapy (40-50Gy) (12). In another study Kitley Et. al, described a case of prostatic cancer with metastasis to penis and causing priapism which was managed successfully by radiotherapy and antiandrogenic medications (16). Similarly, Cante et. al. reported a case of malignant priapism secondary to prostate adenocarcinoma treated with the same combination of androgen deprivation and radiotherapy (17).

There are also other studies have reported the metastasis of the prostate cancer to penis. Nason GJ. Et al. reported a case of a 90-year-old man presented with painless hematuria and increased PSA. The histology of the penile biopsies revealed poorly differentiated carcinoma infiltrating into the dermis. PSA immunohistochemistry was positive, confirming metastatic prostate carcinoma (18). An 83-year-old man with solitary penile metastasis from prostate adenocarcinoma presented with a painless lump at the base of the penis for a few months. He also complained of lower patient urinary tract symptoms. Dedicated penile magnetic resonance imaging (MRI) revealed an enhancing $2.1 \times 1.1 \times 1.3$ right corpus cavernosal nodule along the mid-shaft of the penis. It appeared locally aggressive with infiltration of the enveloping tunica albuginea (19).

Clinical Key Message (Conclusion)

This study presents a case of prostatic adenocarcinoma recurrence manifesting by priapism which was diagnosed and managed successfully. This rare presentation of prostate malignancy relapse is a reminder for practitioners and urologists that priapism can be caused by malignancies. It also can be an educational noting that the combination of radiotherapy and androgen deprivation can save the patient from stressful surgeries such as penectomy. Therefore, these surgeries can be saved as the last resort in malignant priapism.

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Consent Declaration: The patient provided informed consent for the publication of this report, and the center's ethical policy performed the procedure.

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