CASE REPORT

Unilateral testicular metastasis of low PSA level prostatic adenocarcinoma

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SUMMARY

A 71-year-old man presented with a firm left testicle 15 years after being diagnosed with prostatic adenocarcinoma. After the initial diagnosis, the patient underwent a radical prostatectomy. He received radiation therapy 5 years after surgery for a slightly elevated prostate-specific antigen (PSA) level and a negative metastatic workup. Eight years later, the patient's PSA rose to 3.38 ng/mL and he was started on intermittent Lupron therapy. It was then 3 years later when the patient discovered the testicular mass. At that time, his PSA level was 2.98 ng/mL. He underwent a left orchiectomy which demonstrated metastatic prostatic adenocarcinoma. Currently, the patient is doing well 3 months postorchiectomy and is now receiving continuous testosterone suppression therapy. This case report discusses the unique modes of spread from the prostate to the testicle as well as the importance of follow-up care in patients with prostate cancer, especially in the setting of low PSA levels.

BACKGROUND

Prostatic adenocarcinoma is the most commonly diagnosed cancer in men. The average age at diagnosis is 68 years. Metastatic prostatic adenocarcinoma most commonly spreads to the pelvic lymph nodes and the bones. Metastasis to the liver, lungs and other visceral organs is less common.¹ Testicular metastasis of prostatic adenocarcinoma is a very rare event with an incidence at autopsy of 0.02–2.5%.² Even with this low incidence, prostate cancer is the most common type of cancer to metastasise to the testes.³ The metastatic spread of prostate cancer to the testes has been accepted as a sign of advanced disease.

The serum prostate-specific antigen (PSA) is a protein released by prostate cells. Following PSA levels is a way to screen for prostate cancer. After definitive treatment for prostate cancer it also serves as a predictor of outcome to see how the disease responds to treatment.⁴ In regard to monitoring outcome in prostate cancer, it is important to note that disease may progress despite low or undetectable PSA levels.

In this report we discuss a case of prostatic adenocarcinoma seen in a 55-year-old man which metastasised to the left testicle 15 years later. This rare location of metastasis suggests unique possibilities of spread from the prostate to the testicle. This report also demonstrates a metastatic case of low PSA level prostate cancer which is usually a sign of a more aggressive disease.

CASE PRESENTATION

A 55-year-old Caucasian man presented to his primary care provider with haematuria in April 1999. The patient was referred to a urologist for further evaluation. On examination, the patient was noted to have a boggy and enlarged prostate. The patient underwent a biopsy of the right and left lobes of the prostate which revealed prostatic adenocarcinoma with a Gleason score 3+3=6. The patient subsequently had a total suprapubic prostatectomy and nodal dissection in June 1999. The pathology revealed bilateral moderately differentiated prostatic adenocarcinoma Gleason score 3+3=6, extensively involving the left lobe with extension into the capsule with a positive proximal margin and a negative distal margin. Along with the prostate, a left iliac node, left obturator node, right obturator node, bladder neck tissue, and the left seminal vesicle were removed and all tested negative for prostatic adenocarcinoma. A bone and CT scans of the abdomen and pelvis carried out after surgery were negative for residual or metastatic disease. The patient did well postoperatively and his haematuria resolved.

The patient continued to do well until August 2004 when his PSA level rose to 0.22 ng/mL. The patient underwent a bone and CT scans of the abdomen and pelvis which were negative for metastatic disease. The radiation oncologist was concerned about a local recurrence given the long interval to the rise of his PSA as well as the positive margin prostatectomy so the patient underwent external beam radiation therapy. The patient continued to do well after radiation until September 2012 when his PSA rose to 3.38 ng/mL. This rise in PSA level suggested recurrent disease. After a negative metastatic workup, the patient was started on intermittent testosterone suppression therapy in the form of Lupron injections. He received his first 6-month dose of Lupron September 2012 when his PSA rose to 3.38 ng/mL. The PSA measured in March 2013 dropped to 0.75 ng/mL. The second 6-month dose of Lupron was given in September 2013 when his PSA rose to 2.47 ng/mL. The PSA measured in March 2014 dropped to 0.49 ng/mL. These drops in PSA mark a response to testosterone suppression therapy. The third 6-month dose was given in September 2014 when his PSA rose to 2.98 ng/mL. It was at this time that the patient noted the development of a left testicular mass.

The patient, then 71 years of age, was seen by his urologist who confirmed the presence of a left testicular mass. An ultrasound was performed which revealed that the left testicle was within

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normal limits for size but contained a complex solid and cystic vascular intratesticular mass measuring 3.1×3.3×2.4 cm with surrounding fluid and debris. The right testicle was unremarkable. β-Human chorionic gonadotropin and α-fetoprotein levels were negative.

TREATMENT

The patient underwent a left testicle orchiectomy in October 2014 which revealed metastatic prostatic adenocarcinoma with a Gleason score of 3+4=7 nearly completely replacing the normal testicular tissue and focally extending into the epididymis (figure 1). Confirmatory PSA and P504 stains were positive (figure 2). The spermatic cord was unremarkable.

OUTCOME AND FOLLOW-UP

The patient is currently alive and doing well. The PSA level obtained post-unilateral orchiectomy was 0.34 ng/mL and fell to 0.17 ng/mL 1 month later. The patient's treatment was switched from intermittent testosterone suppression to continuous testosterone suppression in the form of Lupron injections. The patient was also presented the option of a right testicle orchiectomy.

Even with the documentation of metastatic disease to the testicle, the patient's PSA level never rose above 3.38 ng/mL throughout the entire 15 years post-total prostatectomy. It is important to note that the patient's testicular metastasis developed during the 6 months that he was off testosterone suppression therapy.

DISCUSSION

This case report demonstrates a very rare and interesting presentation of metastatic prostatic adenocarcinoma. Not only is the site of metastasis unique, but so is the long length of time between initial diagnosis of the low PSA level prostatic adenocarcinoma and the finding of metastatic disease. This is curious because low PSA level prostatic adenocarcinoma tends to be more aggressive in nature.

One of the aspects that make this case unique is the testicular metastatic location of the prostate cancer. Prostate cancer most commonly metastasises to the lymph nodes, bones, lungs, liver, bladder or brain.¹ Testicular metastasis is very rare and usually discovered incidentally at autopsy or following a bilateral orchiectomy for hormonal management of prostate cancer.⁵ This location of disease suggests unusual routes of spread from the prostate such as retrograde venous extension, arterial embolism,

Figure 2 Low-power view of confirmatory prostate-specific antigen stain demonstrating the presence of prostate tissue in the testicular mass.

retrograde lymphatic extension, or direct extension through the lumen of the vas deferens.⁶

Another aspect of this case that is unique is that the patient's PSA levels remained relatively low throughout the entire course of his prostate cancer and metastatic disease. In regard to this topic, a study was completed at the University of Texas M. D. Anderson Cancer Center in 2006 to investigate cases of prostate cancer with disease progression despite low or undetectable PSA levels. In the study, three theories regarding the advancement of low PSA level prostate cancer were explained. All three theories concentrated on atypical histological variants. They are as follows—a proliferation of cell lines that do not have the capacity to produce PSA such as small cell or neuroendocrine tumours, poorly differentiated prostate cancer cells that have lost their ability to express PSA and a stem cell origin of prostate cancer which evolves to produce an undifferentiated small cell variant.⁴ Although it is unknown whether the pathology from this specific case report demonstrated these histological variants, these theories may have implications for future cases in regard to the prognosis of low PSA level advanced prostate cancer and its potentially more aggressive nature.

In conclusion, this case report highlights the need for continued monitoring of patients with a history of prostate cancer for local recurrence as well as metastatic disease. It also shows the importance of detecting metastatic disease to rare locations such as the testicle especially in the setting of low PSA levels.

Figure 1 Low-power view of the testicular mass showing atrophied seminiferous tubules on the right and invasion by prostate tissue on the left.

Learning points

- ▶ Testicular metastatic prostatic adenocarcinoma is a rare finding and suggests unique routes of spread such as retrograde venous extension, arterial embolism, retrograde lymphatic extension or direct extension through the lumen of the vas deferens.
- ▶ Low or undetectable PSA level prostate cancer which has metastasised may suggest a more aggressive form of disease.
- ▶ This case demonstrates the importance of close follow-up care including PSA level measurements and the physical examination even when 15 years has passed from initial diagnosis.

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