Metastatic prostate cancer found within the hernia sac contents is a rare clinical manifestation. We report a 64-year-old male patient who presented with rare clinical features of prostate cancer. A focal metastasis of prostate cancer was incidentally found in an incised inguinal hernia sac 5 years after radical prostatectomy. The serum prostate specific antigen (PSA) level remained undetectable (< 0.01 ng/mL) prior to herniorrhaphy without any adjuvant therapy. In addition, serial studies performed right after herniorrhaphy disclosed no evidence of overt clinical metastasis. However, his serum PSA level started rising 12 months later and bladder invasion as well as a mass in the cul-de-sac was identified subsequently. The serum PSA level was 2.45 ng/mL at his latest visit, which was 5 years after herniorrhaphy. This case implies that physicians should be more alert in patients with a low preoperative serum PSA level during the period of follow-up. Both serum PSA and digital rectal examination may be important in patients with low preoperative PSA level after radical prostatectomy. [J Formos Med Assoc 2007;106(5):397–399]

Key Words: prostate adenocarcinoma, inguinal hernia, metastasis, prostatic specific antigen

Case Report

A 64-year-old man presented with painless gross hematuria in April 1996. The digital rectal examination revealed a hard nodule at the left lobe of
the prostate and needle biopsies revealed a Gleason score 3+4=7 adenocarcinoma. The initial serum PSA level was 1.5 ng/mL (Abbott Architect system, USA). He then underwent radical retropubic prostatectomy in May 1996. Pathologic grading and staging were Gleason score 3+4=7 and pT3bN0M0 (left seminal vesicle invasion and negative surgical margins). At 2-month postoperative follow-up, serum PSA was undetectable (<0.01 ng/mL) and adjuvant therapy was hence not offered. Follow-up visits were requested every 3 months postoperatively. He remained asymptomatic with an undetectable serum PSA level (<0.01 ng/mL) until April 2001, when he presented with a right inguinal hernia and herniorrhaphy was performed. The hernia was an indirect type with no adhesion of bowel contents. Pathologic examination of the transected sac incidentally disclosed a small area with nests of neoplastic glands within the fibroconnective tissue (Figure 1). These tumor cells were immunoreactive to PSA (Figure 2). He then underwent a thorough metastatic survey, which did not reveal any evidence of osseous, visceral or lymph node metastases. Although the serum PSA level still remained undetectable after operation, it became detectable 12 months later and elevated insidiously to 0.51 ng/mL 28 months following herniorrhaphy. Then we added anti-androgen therapy with cyproterone acetate (50 mg) three times daily. Serial imaging studies still revealed no evidence of clinical metastasis. The following nadir PSA level was 0.11 ng/mL. He presented to us with an episode of gross hematuria in May 2005 (60 months following herniorrhaphy). Cystoscopic examination disclosed a tumor at the left lateral wall of the urinary bladder and pathologic examination of the resected tissue revealed metastatic prostate adenocarcinoma. Subsequent computed tomography showed a soft tissue nodule, about 2.5 cm in size in the cul-de-sac region. There was no evidence of osseous or visceral metastasis. After a combined anti-androgen (cyproterone acetate) and salvage external beam radiotherapy (60 Gy) to the true pelvis, the serum PSA level declined from 2.8 to a nadir level of 0.1 ng/mL 6 months later. However, the serum PSA level started to rise afterwards regardless of continuing anti-androgen therapy. His serum PSA level was 2.45 ng/mL and testosterone level was 2.62 ng/mL at the latest visit, which was exactly 5 years after herniorrhaphy and 10 years after radical prostatectomy.

**Discussion**

The finding of metastatic prostate cancer in an incised hernia sac is extremely rare and the reported cases presented either with apparently abnormal finding in the hernia contents or with very high serum PSA level.3 Our case is unique as the serum PSA level was undetectable when hernia metastasis was found and no overt metastasis to other

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**Figure 1.** A small area of neoplastic glands (arrow) is found in the hernia sac tissue (hematoxylin and eosin, 40×).

**Figure 2.** Strong prostate specific antigen (PSA) cytoplasmic immunoreactivity is demonstrated in the metastatic prostate adenocarcinoma (ABC method, 400×).
Metastatic prostatic adenocarcinoma in an inguinal hernia sac

organs could be detected. Furthermore, unlike the common thought that the discovery of a metastatic tumor in an inguinal hernia sac suggests advanced disease and a short life expectancy,¹ our patient remained alive 5 years after herniorrhaphy.

The route of metastasis in this case is unclear. Presumably, the tumor might have metastasized to the hernia sac via hematogenous dissemination, which would suggest that other sites of metastases might also have existed but remained occult. As the inguinal hernia developed 5 years after radical prostatectomy, it is less likely that the tumor cells were spilled over the peritoneum during radical prostatectomy or had reached the hernia sac via localized spread.⁶,⁷

Distant metastasis from prostate cancer after radical surgery accompanied by undetectable PSA is also very rare and a possible explanation for this phenomenon remains speculative.⁵,⁸,⁹ We could not exclude the possibility of histologic and biochemical dedifferentiations, given the low preoperative serum PSA level (1.5 ng/mL) and high grade (Gleason score, 3+4=7) in our case. It is also likely that the small occult lesion found within the hernia sac might not be able to produce enough PSA to reach a detectable serum level. The prognosis of such cases is not predictable based on the limited reported experience.¹⁰ Our patient remained alive 10 years after radical prostatectomy.

In summary, our case is the first one of metastatic prostate cancer found in the hernia sac with an undetectable serum PSA level. Whatever the mechanism accounting for this finding, a detectable serum PSA level was found eventually and overt distant metastasis developed. Apart from its rarity, the case implies that physicians should be more alert in patients with a low preoperative serum PSA level during the period of follow-up, as even a minor elevation of serum PSA level may indicate a significant distant metastasis. The follow-up protocol for patients who have undergone radical prostatectomy includes not only serum PSA but also digital rectal examination. For patients with low preoperative PSA level, serum PSA alone is not an ideal tool for active surveillance.

References