Case Report

Urosymphyseal fistula development following treatment for radiation-induced urethral stenosis in three patients with prostate cancer

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Abbreviations & Acronyms
DVIU = direct vision internal urethrotomy
EBRT = external beam radiotherapy
EPA = excision and primary anastomosis
HDR-BT = high dose rate brachytherapy
IMRT = intensity modulated radiation therapy
LRP = laparoscopic radical prostatectomy
MRI = magnetic resonance imaging
PBO = pubic bone osteomyelitis
PC = prostate cancer
RT = radiation therapy
SPT = suprapubic tube
USF = urosymphyseal fistula
VUAS = vesicourethral anastomosis stenosis

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Introduction: Urosymphyseal fistula is a rare and devastating complication that develops after radiation therapy for prostate cancer and is often triggered by the treatment of radiation-induced urethral stenosis. Here, we report our experience with urosymphyseal fistulas in three patients with prostate cancer.

Case presentation: Three patients with prostate cancer developed urethral stenosis after radiotherapy. The management of urethral stenosis was suprapubic tube placement in case 1, direct vision internal urethrotomy in case 2, and excision with primary anastomosis in case 3. All patients presented with severe suprapubic or thigh pain or both. Urosymphyseal fistulas were detected on magnetic resonance imaging. Conservative treatment was unsuccessful, and all patients required debridement of the necrotic pubic symphysis and simple cystectomy. In cases 1 and 2, ileal conduit urinary diversion was performed.

Conclusion: Urologists need to be aware that urosymphyseal fistulas can occur in irradiated patients with prostate cancer, especially after urethral stenosis treatment.

Key words: prostate cancer, radiation therapy, surgical management, urethral stenosis, urosymphyseal fistula.

Keynote message

- USF is a rare and devastating complication after RT for PC.
- It should be noted that USF is often triggered by treatment for radiation-induced urethral stenosis.
- Conservative treatment is ineffective and requires surgical intervention, primarily debridement of the necrotic pubic bone, cystectomy, and urinary diversion.

Introduction

USF, a rare and devastating complication of RT for PC, is a fistula extending from the urinary tract to the pubic symphysis.[1] Extravasation of urine from a damaged urinary tract can trigger fistula formation.[2] Therefore, treatment of radiation-induced urethral stenosis, such as through DVIU and urethral dilation, often leads to USF development.[1–3] USF causes local tissue inflammation and destruction, resulting in PBO and recurrent abscess formation. Patients with USF present with pelvic and suprapubic pain, chronic infections, and gait disturbance.[1] Conservative management, such as antibiotic administration, is generally ineffective; surgical interventions, including debridement of the necrotic pubic symphysis and cystectomy with urinary diversion, are often required.[1] Here, we report our experience with the surgical management of three patients with USF.

Case report

Case 1

A 78-year-old man, who underwent EBRT for PC 15 years ago, presented with synchronous urethral stenosis at the penobulbar urethra and bulbomembronous junction (Fig. 1a; Table 1).

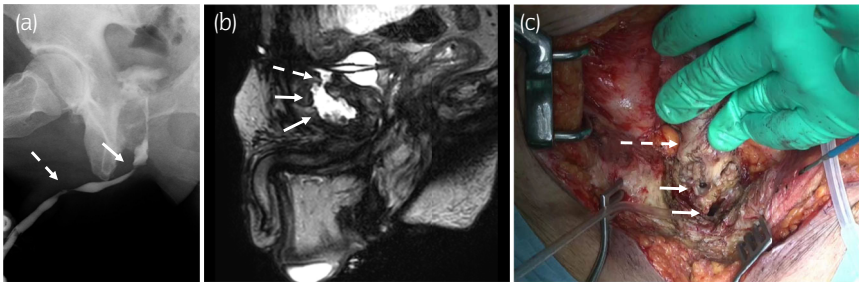

Fig. 1 Radiological and operative findings of case 1. (a) Retrograde urethrography showed synchronous urethral stenosis at the penobulbar urethra (dotted arrow) and the bulbomembronous junction (arrow). (b) T2-weighted MRI in the sagittal plane showed a USF from the SPT tract (dotted arrow) to the pubic symphysis (arrows). (c) Intraoperative finding of USF arising from the SPT tract (dotted arrow) to the pubic symphysis (arrows).

Table 1 Patient characteristics

	Case 1	Case 2	Case 3
Age	78	78	72
Type of treatment for PC	EBRT	LRP Salvage RT	IMRT and HDR-BT
Time from radiation to onset of USF	16 years	5 years	14 years
Symptoms related to USF	Suprapubic pain Thigh pain Gait disturbance	Perineal pain Hip and thigh pain	Suprapubic pain Thigh pain
Location of urethral stenosis	Penobulbar Bulbomembranous	VUAS	Bulbomembranous
Location of fistula in the urinary tract	SPT tract	Vesicourethral anastomosis site	Anastomosis site of EPA
Type of stenosis treatment	Repeat dilations SPT placement	Repeat DVIUs	DVIU Dilation EPA
Time from stenosis treatment to USF	16 months	7 months	5 months
Management	Simple cystectomy Ileal conduit Debridement of pubic bone Omental flap interposition	Simple cystectomy Ileal conduit Debridement of pubic bone Omental flap interposition	Debridement of pubic bone
Outcome	Symptom free	Symptom free	Symptom free

Urethral dilations were performed repeatedly without improvement. Subsequently, a SPT was placed due to urinary retention and uncontrollable hematuria resulting from concurrent radiation cystitis. Sixteen months after the SPT placement, the patient complained of severe suprapubic pain and gait disturbance. PBO, peripubic abscess, and USF arising from the SPT were detected on MRI (Fig. 1b). Although antibiotics were administered for 6 months, the abscess extended to the right thigh, and his symptoms worsened. A simple cystectomy and creation of an ileal conduit were performed in combination with debridement of the necrotic pubic symphysis and coverage with an omental flap interposition. The USF was connected to the SPT tract and pubic symphysis (Fig. 1c). Pelvic pain improved in the early postoperative period, and there was no recurrence 2 years postoperatively.

Case 2

A 78-year-old man underwent salvage RT for local PC recurrence following LRP (Table 1). Repeated hematuria due to radiation cystitis was frequently observed. VUAS was identified 2 years after RT (Fig. 2a), and DVIU was performed using a hot knife at the 3 and 9 o'clock positions.[4] Six months after DVIU, perineal discomfort and left hip pains were observed. PBO and USF extending from the

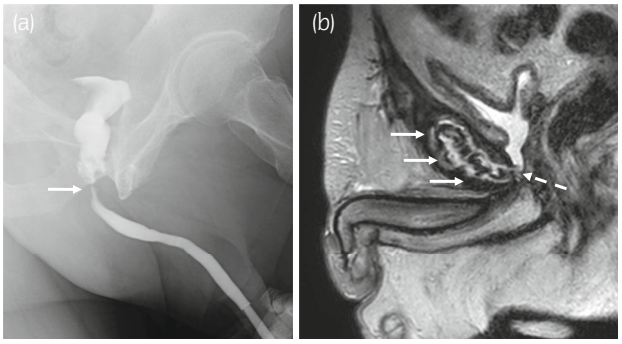

Fig. 2 Radiological findings of case 2. (a) Retrograde urethrography showed VUAS (arrow). (b) T2-weighted MRI in the sagittal plane showed a fistula arising from the vesicourethral anastomosis site (dotted arrow) to the pubic symphysis (arrows).

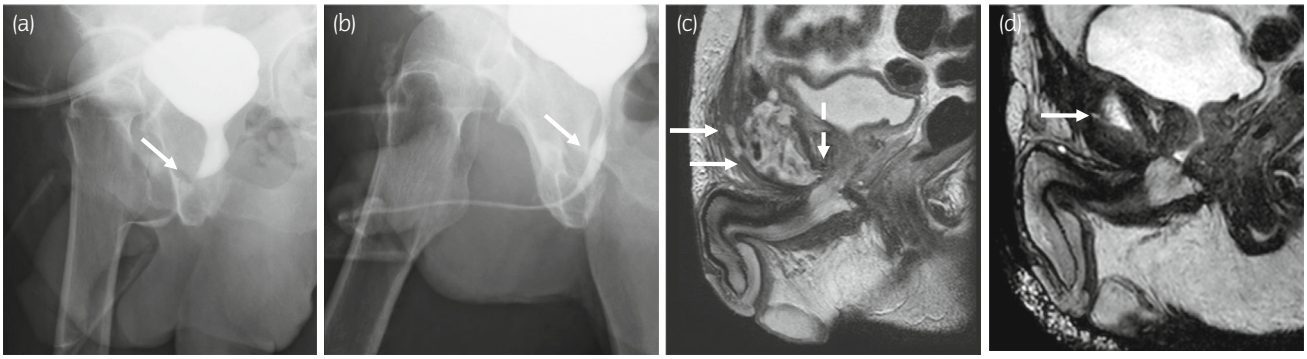

Fig. 3 Radiological findings of case 3. (a) Retrograde urethrography showed urethral stenosis at the bulbomembranous junction (arrow). (b) Retrograde urethrography after EPA showed a wide urethral lumen without extravasation. (c) T2-weighted MRI in the sagittal plane showed USF from the anastomosis site (dotted arrow) to the pubic symphysis (arrows). (d) T2-weighted MRI in the sagittal plane after the debridement of necrotic pubic bone (arrow).

VUAS site to the pubic symphysis were detected on MRI (Fig. 2b). After failed conservative management with antibiotics administration for 4 months, simple cystectomy and creation of an ileal conduit were performed in combination with debridement of the necrotic pubic symphysis and coverage with an omental flap. No recurrence was observed 18 months postoperatively.

Case 3

A 72-year-old man, who had received IMRT and HDR-BT for PC 13 years ago, had a bulbomembranous urethral stenosis that was successfully treated with EPA (Fig. 3a,b; Table 1). Five months after EPA, lower abdominal and pelvic pain were observed, and PBO was detected on MRI (Fig. 3c). USF arose from the anastomosis site to the pubic symphysis. Although antibiotics were administered for 6 months, the chronic infection was uncontrollable. Finally, the necrotic pubic symphysis was debrided. The fluid collection remained in the pubic symphysis (Fig. 3d), but the fistula arising from the urethral anastomosis and the pelvic pain disappeared spontaneously. Two years after debridement, we found no worsening of the symptoms.

Discussion

RT is a standard treatment for localized PC and a salvage treatment for local recurrence after prostatectomy, but it carries a 2–5% risk of urethral stenosis, with increasing risk over time.[5,6] A recent systematic review found that transurethral treatment for urethral stenosis triggered USF in 83% of patients with irradiated PC.[1] Our report further revealed that USF can result from not only transurethral treatment but also SPT placement and urethroplasty, indicating that any urinary tract opening poses a risk for USF in irradiated PC patients.

USF symptoms vary with fistula spread and include severe pain in the suprapubic, groin, and thigh areas.[1] Diagnosis delay due to the atypical nature of USF may necessitate prolonged opioid use.[7] MRI is crucial, confirming USF in 95% of cases.[1] MRI can depict minor fistulas, concurrent PBO, and surrounding fluid collection.[1] Therefore, avoiding hesitation when using MRI for early diagnosis is essential.

Cases 1 and 2 were selected for cystectomy and urinary diversion because urethral stenosis was challenging to repair, and bladder compliance was poor due to radiation cystitis. In contrast, Case 3 was selected for primary repair (debridement of necrotic tissue and USF closure with soft tissues) because EPA had fixed the stenosis, and the bladder capacity was adequate. Although a small amount of fluid was collected from the pubic symphysis, the USF resolved, and the symptoms were well controlled.

In our study, all patients showed symptomatic improvement without recurrent symptoms in the early postoperative period. Long-term infection and persistence of symptoms can impair quality of life, underscoring the importance of early diagnosis and timely surgery.

Conclusion

Patients with symptoms suggestive of USF who have undergone RT for PC should maintain a high suspicion. MRI should be the diagnostic test of choice to confirm the diagnosis and surgical intervention should be considered from the onset of the condition.

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None.

Author contributions

Kazuki Takekawa: Data curation; writing – original draft. Akio Horiguchi: Conceptualization; data curation; supervision; writing – original draft. Kenichiro Ojima: Conceptualization; data curation; supervision. Masayuki Shinchi: Data curation; supervision. Yusuke Hirano: Data curation; supervision. Yoshiyuki Furukawa: Data curation; supervision. Keiichi Ito: Supervision. Ryuichi Azuma: Data curation; supervision.

Conflict of interest

The authors declare no conflict of interest.

Approval of the research protocol by an institutional reviewer board

Not applicable.

Informed consent

Informed consent was obtained from the patients for publication of this case report.

Registry and the registration no. of the study/trial

Not applicable.

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