Case Report

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Urethral Tumors A Report of 6 Cases

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INTRODUCTION

Primary urethral carcinoma is a very rare cancer in men and generally occurs at the 5th decade of life.⁽¹⁾ Its most common types are squamous cell carcinoma (SCC), transitional cell carcinoma (TCC), and adenocarcinoma, in order of prevalence. The incidence of different histologic subtypes of these tumors vary at each part of the urethra.⁽²⁾ The incidence rate of urethral TCC after radical cystectomy is 8% to 10% and its standard treatment is total urethrectomy.⁽¹⁻³⁾ Some cases of late recurrence in the meatus have been reported following subtotal urethrectomy. This has put the value of this technique into question during the recent years.(3-5)

Between March 2005 and November 2005, 3 cases of primary urethral

tumors were treated at Modarres hospital. Also, during this period, 3 cases of tumor recurrence in the urethra after radical cystectomy were diagnosed, in 2 of which, the patients had previously undergone subtotal urethrectomy and the recurrence occurred in the glandular part of the urethra. We report these 6 cases in our study.

CASE REPORT

Case 1

An 82-year-old man presented with obstructive urinary tract symptoms and bloody discharge from the urethra. On physical examination, a pedunculated tumor sized 1×1 cm was seen in the fossa navicularis which could be protruded by pressure. The distal part of the penis was completely



Figure 1. In Case 1, a pedunculated tumor was found in the fossa navicularis which could be protruded by pressure. The distal part of the penis was completely indurated.



Figure 2. Partial penectomy in Case 1.

indurated and inguinal lymphadenopathy was not detected (Figure 1). Urine cytology revealed atypical and inflammatory cells. Biopsy revealed a grade 3 TCC. Metastatic workup was unremarkable. Also, on cystoscopy, tumoral lesion was not seen in other parts of the urethra.

The patient underwent partial penectomy (Figure 2). The pathologic evaluation of the specimen revealed a grade 2/3 TCC with the involvement of the submucosa but with free margins. Three months postoperatively, the patient was symptom-free. On cystoscopy, meatus was completely open and no recurrence was detected in the urethra and the bladder. Urine cytology result was negative for malignancy.

Case 2

A 44-year-old man was referred with perception of a mass in penoscrotal junction of the urethra since 1 year earlier. The patient also complained of swelling and pain in the external genitalia during the previous 3 months. Ulcerative gangrenous necrotic lesions were found on the penis and the scrotal skin, with a malodorous purulent discharge due to bacterial superinfection. The penile shaft was attached to the body only by a few skin bridges, and the penis was actually auto-amputated. Bilateral firm inguinal lymph nodes were detected, fistulated to the skin on the left side (Figure 3).

A biopsy had been taken from the urethral mass and the pathology report was moderately differentiated



Figure 3. A bilateral firm inguinal lymphadenopathy fistulated to the skin was detected in Case 2.

invasive SCC. After releasing the skin bridges, total penectomy was performed. Biopsies from lymph nodes revealed invasive SCC.

Case 3

An 83-year-old diabetic man with irritative lower urinary tract symptoms and obstruction was referred to our clinic. On urethroscopy, papillary and sessile lesions were seen in the penile, bulbous, and prostatic urethra. No tumors were found in the bladder and the intravenous urography did not show any abnormality. Biopsy of the tumoral lesions revealed a grade 2 TCC with invasion to the fibrous stroma.

Radical surgery was not performed due to the poor general condition of the patient, and bilateral orchiectomy was performed.

Case 4

A 52-year-old man presented with a history of radical cystectomy and bilateral cutaneous ureterostomy in 2000. The patient had experienced bloody discharge from the urethra 2 years after the surgical operation. Following the recurrence of the tumor in 2003, subtotal urethrectomy (with sparing the glandular urethra) had been performed. Pathologic report was grade 2 TCC. In 2004, the patient had undergone left nephroureterectomy due to several tumoral lesions.

The patient returned in 2005 with bloody discharge and clot passage. On physical examination, a 0.5×0.5 -cm papillary tumor was seen in the fossa navicularis (Figure 4). Incisional biopsy was



Figure 4. A papillary tumor was detected in the fossa navicularis in Case 4.

performed on the lesion and a papillary TCC was reported. The lesion and a part of the glans of penis were excised.

Case 5

An 81-year-old man presented with bloody discharge from the meatus. The patient had a history of radical cystectomy and ileal pouch due to muscle-invasive bladder tumor (grade 2 TCC) about 9 years earlier. On urethroscopy, multiple papillary tumors were seen in the proximal bulbar and membranous urethra. Total urethrectomy was performed and a grade 3 TCC was reported.

Case 6

A 46-year-old man had undergone radical cystectomy and urethrectomy due to bladder cancer (TCC). One year afterwards, the patient referred with pain and purulent discharge from the ulcerated and necrotic lesions on the penile glans. On physical examination, tender, firm, bilateral lymph nodes were detected in the groins. Pathologic examination of the lesions revealed TCC.

DISCUSSION

Primary Urethral Carcinoma

Urethral carcinoma is a rare condition especially in men,⁽¹⁾ and the most common presentations are bloody discharge from the meatus, urinary obstructive symptoms, and palpable masses in the anterior urethra. These tumors are usually detected in the bulbomembranous, penile, and prostatic urethra (60%, 30%, and 10%, respectively).⁽¹⁻³⁾ In men, 80% of the tumors are SCC, while only 15% and 5% of these tumors are TCC and adenocarcinoma, respectively.⁽¹⁻³⁾ The histologic characteristics of a tumor is different depending on its anatomic location. In the posterior urethra, most detected tumors are TCC, while in the anterior part they are mostly SCC.⁽²⁾

Transitional cell carcinoma is a panurothelial disease. Thus, before regarding it as a primary urethral tumor, it is necessary to rule out the involvement of other parts of the urothelium (especially by carcinoma in situ).

Most of the tumors present in advanced stages as changes in voiding habits. Therefore, attention to

the possibility of urethral tumor in all patients with urinary tract symptoms is crucial. A complete history (focusing on hematuria, bloody discharge from the urethra, or perception of a mass in the urethra), physical examination (palpation of penis and corpus cavernosum up to the perineum), and paraclinical studies should be performed in all of these patients.

In our first case, despite all diagnostic efforts, we could not detect any finding indicative of the involvement of the other urothelial parts. It could be concluded that the glandular urethral TCC of this patient was most probably isolated. This tumor is noticeable and very rare among the primary urethral tumors, because only 3% of all urethral tumors in men are TCC of the penis.⁽¹⁻³⁾

Large tumors in the ventral part of the urethra can cause a periurethral mass or fistula. Of other manifestations of urethral tumors are penile erosion, induration, and penile swelling.⁽²⁾ But, auto-amputation of the penis due to the urethral cancer, as seen in our second case, is an extraordinary presentation, and we could not find any similar report in the literature.

Urethral TCC after Radical Cystectomy

The incidence of urethral TCC after radical cystectomy is 8% to 10% and occurs 1.5 to 2.5 years therafter. Most of the tumors manifest within 5 years.⁽³⁾ Risk factors associated with the tumor recurrence are multifocal primary tumor, carcinoma in situ, upper tract TCC, and the involvement of the bladder neck and prostatic urethra.⁽⁵⁾ None of these factors are absolute contraindications for urethral preservation during radical cystectomy. It has been shown that taking frozen sections from the urethral margin during the operation is the most sensitive method for prediction of the tumor recurrence.^(6,7)

In some studies, it has been recommended to perform transurethral biopsy to evaluate the involvement of the prostate before radical cystectomy,^(5,8) while in others, it has been emphasized that even prostate involvement cannot be a preclusion for orthotopic diversion if the margins are negative in frozen sections.^(6,7,9)

In the management of the patients who are candidates for radical cystectomy, a correct selection for functional or dry urethral preservation is of utmost importance. Also, it is essential to monitor the urethra for the rest of the patients' life. Physical examination, washing cytology, and urethroscopy are recommended at least annually. Attention to the symptoms of tumor recurrence such as bloody discharge from the urethra, hematuria, and changes in voiding habits are necessary.⁽³⁾

The ideal treatment for tumor recurrence in the urethra is total urethrectomy. Attempts for subtotal urethrectomy (with meatal sparing) for facilitating the use of prosthesis has been accompanied by a high rate of tumor recurrence in glandular part of the urethra.^(3,4,10,11) Schellhammer and Whitmore observed involvement of this part of the urethra in 27% of the patients who had undergone subtotal urethrectomy following the recurrence of the tumor after cystectomy.⁽¹²⁾ In a cohort study on 1054 subjects with a follow-up more than 10 years, 2 patients have been diagnosed with late recurrence of the tumor in the meatus (3 and 11 years later), both with a history of subtotal urethrectomy.⁽³⁾ Endoscopic management with or without intraurethral treatments has been attempted in some cases of superficial TCC that recur after orthotopic diversion.^(3,13,14)

Cases 4 and 6 were recurrences of tumor in the preserved urethras 2 and 1 years after the urethrectomy. In Case 6, the recurrence was extensive and the corpus cavernosum and the penis were also involved, which made curative treatments impossible. In Case 5, late recurrence of TCC (9 years after radical cystectomy) was observed in the posterior urethra. By reviewing the biologic models of oncogenesis, it can be concluded that late recurrences are mostly because of field changes in the whole urothelium rather than derivatives of primary tumor cloning.

In summary, it seems that subtotal urethrectomy following radical cystectomy for facilitating the later use of prosthesis is accompanied by a higher recurrence rate in the glandular part of the urethra. The authors recommend total urethrectomy as the standard treatment in these patients, especially in Iran where prosthesis is not commonly used.

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