Transitional cell carcinoma (TCC) can develop as a multifocal tumor in the urinary system, especially in the bladder. Here, we report a 69-year-old man who had undergone transurethral resection for bladder tumor and had urethral recurrence that presented as a perineal mass 2 years after treatment. However, he had obtained normal cystoscopy, ultrasonography and computed tomography results at follow-up examinations.

**Key Words:** bladder tumor, perineal mass, transitional cell carcinoma, urethral tumor

Transitional cell carcinoma (TCC) can develop as a multifocal tumor in the urinary system, especially in the bladder [1]. The possible causes of synchronous and asynchronous anterior urethral TCC development in bladder TCC patients, who have been treated with the orthotopic new bladder procedure, have been discussed in several reports in the literature [2–5]. Anterior urethral TCC recurrence after cystectomy has been reported in 4–17% of patients. Multifocal cancers, concurrent upper urinary tract tumors, diffuse carcinoma in situ (CIS), involvement of the bladder neck or trigone, involvement of the prostatic urethra or deep prostatic invasion, and positive urethral margins on intraoperative frozen sections are associated with increased risk of urethral recurrence [2,4–6]. Although prophylactic urethrectomy is recommended in these conditions, there are still discrepancies among the studies in the literature. Tobsu et al detected anterior urethral TCC in 10.6% of their patients that had undergone radical cystectomy for bladder TCC [7]. In another study, they detected TCC in bulbar urethra in 19% of patients who had been treated with cystourethrectomy in the presence of diffuse bladder CIS, whereas no bulbar urethral tumor was seen in patients without diffuse CIS [8].

In our literature search, we found case reports about the management approaches for urethral tumor recurrence after radical cystectomy. However, we could not find a clear approach for the treatment of patients who refuse radical cystectomy. The aim of the study was to evaluate treatment options and present the clinical features of an invasive bladder tumor (T2a, G3) in a patient who refused advance treatment options for invasive bladder tumor, and had urethral recurrence 2 years after transurethral resection.

**CASE PRESENTATION**

A 69-year-old man underwent transurethral resection for a bladder tumor in 2006. Cystoscopy during transurethral resection showed that the tumor was 3–4 cm in diameter, localized in the left lateral wall and projected into the lumen in a papillary formation. The tumor was resected with a 1-cm margin on non-tumor tissue. Pathological evaluation revealed
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T2a, G3 TCC. A single dose of mitomycin C was administered just after the operation. The patient refused all treatment modalities for invasive bladder tumor. Therefore, he was subjected to follow-up examination every 3 months. The patient presented to us with obstructive lower urinary tract symptoms (LUTS) in June 2008. Physical examination revealed a perineal mass that the patient had recognized during the previous 2 months, which had grown rapidly since then. This perineal mass caused obstructive LUTS and the patient had a history of bladder tumor, therefore, we suspected that it was a metastatic or recurrent tumor. Whole abdominal and pelvic computed tomography (CT) revealed a urethral mass of 3.5 × 2.0 cm (Figure 1A). Radiological examination and cystourethroscopy did not reveal any other malignancies. Thereafter, partial urethrectomy with perineal urethrostomy were performed (Figure 1B). The patient did not accept total or partial penectomy because he felt that erectile function was the most important issue for him. The tumor was located on the bulbar urethra. Pathological examination revealed pT2 TCC of the bulbar urethra, with dimensions of 3.8 × 2.8 × 2.5 cm (Figure 2). The surgical margin was clear but there was perineuronal invasion. The patient was followed up every 3 months. Generally, the follow-up evaluation at each visit included a physical examination, urinary cytology, ultrasonography, cystourethroscopy and, if necessary, CT, bone scintigraphy and chest radiography. Physical examination, cystoscopy and ultrasound images were normal in follow-up examinations at 3, 6, 9, 12 and 15 months. No pathological findings were detected on total abdominal and pelvic CT scans that were done at 6 months and 12 months. The patient’s family was also persuaded to undergo chemotherapy.

The patient was treated with combination chemotherapy including cisplatin because he had advanced disease. He showed no evidence of metastasis or local recurrence for 16 months after surgery. Postoperative urine cytology was negative. The patient was healthy and free of complications and recurrence at 16 months after the operation.

DISCUSSION

Urethral malignancy accounts for <1% of male urological malignancies. The most common histological type
is squamous cell carcinoma (78%) in the anterior urethra, which is lined by pseudostratified and stratified columnar epithelium, and most distally in the meatus by stratified squamous epithelium. One in five of anterior urethral carcinomas occur in the fossa navicularis. TCC usually occurs in the posterior urethra; mainly in the prostatic portion where the epithelium is transitional and in continuity with the bladder epithelium. TCC is extremely rare in the anterior urethra. Its occurrence in an area that is normally lined by squamous epithelium could be explained by urothelial metaplasia, urogenital tumor metastases, or the presence of ectopic urothelium.

It is also uncommon for recurrent urethral tumor to present as a painful perineal mass. Mass lesions that arise in the perineum of patients who have undergone cystectomy for urothelial carcinoma should raise suspicion of recurrent urothelial carcinoma. Urethral recurrence has been reportedly identified by several symptoms, or by cytological examination. Initial symptoms for urethral recurrence are obstructive LUTS, macrohematuria, penile pain or mass, and urinary incontinence [9,10]. The prognosis of superficial urethral recurrence is better than that of invasive recurrence; therefore, early detection is needed for such urethral recurrence. Retrospective analysis of cystectomy series has revealed a correlation between specific pathological characteristics of bladder TCC and anterior urethral recurrence, including tumor multiplicity, existence of diffuse CIS, involvement of the internal urethral orifice and prostatic urethra, and particularly, invasion into the prostatic ducts or stroma. It has been reported that 4–18% of male patients will develop recurrent urethral TCC in the remnant urethra after cystectomy [4,5–11]. Gowing has found CIS in the urethra in six of 33 autopsies performed on patients who died from bladder cancer [12]. Nieder et al observed urethral recurrence in eight of 218 patients (3.7%) who underwent radical cystoprostatectomy [13]. These studies suggest the treatment procedure that should be applied in the case of urethral spread or recurrence detected during or after cystectomy. However, there are still no clear data for patients with urethral recurrence that develops after transurethral resection for invasive bladder tumor, even though they have normal cystoscopy, ultrasonography and CT results at follow-up. Our patient underwent follow-up examination every 3 months. However, the patient presented to us with obstructive LUTS after 2 years. Perineal mass was detected on physical examination, and urethral recurrence was suspected.

Tumors of the anterior urethra distal to the bulb are best managed by partial or total penectomy with perineal urethrostomy. Local recurrences are rare after such treatment. Treatment failure is a result of an unsuspected inadequate surgical margin or undetected regional lymph node involvement at the time of primary surgery [14]. However, our patient underwent partial urethrectomy with perineal urethrostomy because he did not want total or even partial penectomy. He mentioned that erectile function was the most important factor for him. The reported 5-year survival rate is between 50–66%. It is important to be aware that symptoms of some benign diseases such as urethral stricture, urethritis, prostatitis and prostatic enlargement are also those of urethral malignancy. The development of a stricture in a middle aged or elderly man with no previous history of urethral disease, especially obstruction that bleeds easily, should arouse suspicion of urethral malignancy [15].

REFERENCES