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Case Report

Laparoscopic Radical Cystourethrectomy in a Patient with Adenocarcinoma of the Female Urethral Diverticulum

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Adenocarcinomas arising in the female urethra have been rarely reported. Here we report a case of laparoscopic radical cystourethrectomy with incontinent urinary diversion in a patient with adenocarcinoma in the urethra and bladder. A 60-year-old female presenting with a history of recurrent cystitis and painless hematuria was referred to our facility with voiding difficulty and a urethral mass. Radiologic evaluation showed an enhanced mass in the urethra and bladder neck. Cystoscopic biopsy of the mass in the bladder neck revealed an adenocarcinoma. Laparoscopic radical cystourethrectomy with anterior vaginal wall excision followed by extracorporeal incontinent urinary diversion was performed.

Key Words: *Adenocarcinoma; Laparoscopy; Urethra*

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Primary carcinomas arising in a urethral diverticulum have rarely been reported and account for only 5% of all urethral malignancies [1]. Most of these tumors arise from squamous or transitional cells, and only 20% are adenocarcinomas of unknown origin.

Because of the rarity of such tumors, a treatment strategy has not been established, but radical cystourethrectomy with pelvic lymph node dissection and urinary diversion appears to be the most beneficial modality for non-metastatic disease. Many authors have reported on an open approach; however, we performed laparoscopic radical cystourethrectomy with bilateral pelvic lymph node dissection and anterior vaginal wall excision followed by ileal urinary diversion with a minimal abdominal incision.

CASE REPORT

A 60-year-old female with a history of recurrent cystitis and painless gross hematuria for 3 years was referred to our institution complaining of intermittent urinary retention. The patient had undergone a radical hysterectomy with bilateral salpingo-oophorectomy due to squamous cell type cervical cancer 20 years ago. Physical examination revealed a firm mass on the anterior vaginal wall. Urine cytology was nonspecific. Serum CEA and CA 19-9 were within normal limits. During cystourethroscopy, a papillary growing tumor was found at the bladder neck (Fig. 1). We found no definite mass lesion during urethrography; however, a urethral narrowing was revealed. Magnetic resonance imaging (MRI) showed a urethral mass invading the base of the bladder and the anterior vaginal wall with no pelvic lymph node enlargement. The mass showed homogeneous, low signal intensity on T1-weighted images and inhomogeneous contrast enhancement after gadolinium administration. On T2-weighted images, the mass showed high signal intensity surrounded by a low signal intense rim (Fig. 2).

A fan-shaped four-port laparoscopic transperitoneal approach was performed (one 10 mm port placed at 10 mm above the umbilicus [camera], one 12 mm right pararectal trocar, one 12 mm trocar placed laterally in the right lower quadrant [assistant port], and one 5 mm trocar placed between the left anterior iliac spine and the umbilicus).

Mild adhesion was observed around the ureter. After ureteral dissection, we clamped the ureter close to the ureterovesical junction by using a Hem-o-Lok clip and performed a frozen biopsy. The results of the frozen biopsy were negative and right side pelvic lymph node dissection was performed. Left side ureteral dissection and pelvic lymph node dissection were also performed as right side

maneuvers.

The peritoneum was incised at the vesicovaginal culde-sac and the lateral pedicles of the bladder were clamped by using Hem-o-Lok clips. The bladder and vagina were dissected around carefully with a finger inserted into the vagina because gas leakage was possible via the vaginal opening. A large prostate-like mass was identified in front of the bladder.

We incised the peritoneum along the medial umbilical ligaments and dissected the anterior perivesical space. The urethral mass similar to a large prostate was revealed in front of the bladder and we incised the endopelvic fascia. The dorsal vein complex was coagulated. As my assistant drew the bladder in the cephalic direction and compressed the vestibule to protect against air leakage, I performed a urethrectomy with anterior vaginal resection. The specimen was placed into a specimen retrieval bag for removal. Urethral and vaginal sutures were made with Vicryl 2-0 laparoscopically.

A 5 cm midline incision was made and the specimen was extracted. A 20 cm ileal segment was chosen and an ileal conduit procedure was performed by using an open-assisted technique. The total operative time was 405 minutes, and the estimated intraoperative blood loss was 580 ml.

In the pathologic gross examination, an exophytic friable mass measuring 5.6x5.3 cm was found at the urethra and bladder neck. Clear cell adenocarcinoma was found at the

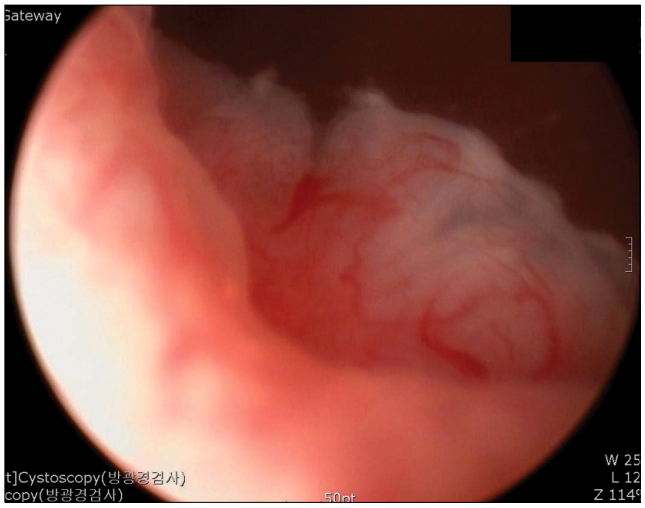

FIG. 1. Cystoscopy showed an exophytic mass at the bladder neck.

FIG. 2. The urethral mass invaded to the bladder neck and anterior vaginal wall, but there was no pelvic lymph node enlargement. Gadolinium-enhanced magnetic resonance imaging (MRI) showed an irregular enhanced mass that invaded the bladder neck (white arrows).

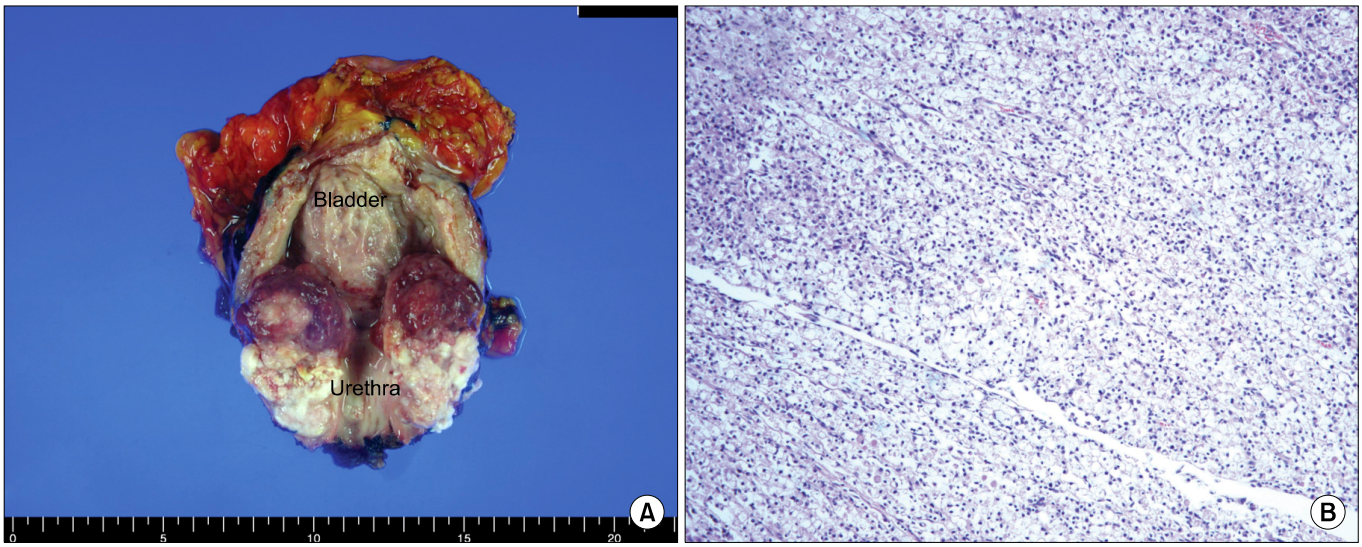

FIG. 3. Gross findings were of an exophytic friable mass at the urethra and the bladder neck, measuring 5.6x5.3 cm. The lesion invaded the periurethral connective tissues (A). Microscopic examination of the tumor (H&E, x40) revealed clear cell adenocarcinoma (B).

urethra and bladder neck, and the tumor had invaded the periurethral connective tissues (Fig. 3). However, all surgical margins and both pelvic lymph nodes were free of tumors. In the immunohistochemical study, the tumor cells showed positive immunoreactivity to CK7 and were negative to CK20, CEA, and CD10. Drop infusion pyelography performed at postoperative day 14 showed no leakage at the uretero-conduit anastomosis sites, and the patient was discharged without complications. After 3 and 6 months with no adjuvant treatment, computed tomography showed no local recurrence or distant metastasis.

DISCUSSION

Urethral carcinomas are uncommon, and make up less than 0.02% of all female carcinomas [1]. Although squamous cell carcinomas are usually identified in female urethral carcinomas, more than half of carcinomas in the urethral diverticulum are adenocarcinomas.

Carcinoma arising in the female urethral diverticulum was first reported by Hamilton and Leach in 1951 [2], and about 100 cases of female urethral carcinoma associated with urethral diverticulum have been reported. Although the histogenesis of urethral adenocarcinoma is controversial, it occurs in the urethral diverticulum and may arise in the paraurethral ducts and Skene glands [3].

Patients usually complain of difficulty in voiding and hematuria, and the most common sign is a palpable suburethral mass [4]. The patient in this case had a history of recurrent urinary tract infections and symptoms of difficulty in voiding.

In magnetic resonance imaging, the normal urethra has a target-like appearance, reflecting 3 histologically distinct layers: an inner lower-intensity layer consisting of urethral mucosa and glands, a middle high-intensity submucosal layer, and an outer low-intensity muscle layer consisting of inner longitudinal muscles and outer striated muscles. Tumors arising from the urethral diverticulum or paraurethral glands may dissect the urethral muscle layer [3]. In this case, the tumor also showed high signal intensity on T2-weighted images and was surrounded by a low-signal-intense ring at the periphery.

Immunohistochemical study can be a helpful tool in identifying the exact type of adenocarcinoma. Park et al reported on the expression of CDX-2 in 60.9% of stomach adenocarcinoma patients and suggested the value of determination of tissue-specific immunohistochemical stains in diagnostic differentiation of adenocarcinomas [5]. Park et al reported that the positive predictive value of CDX-2 (+), CK7 (+), TTF-1 (−), and CK20 (−) in adenocarcinoma was 85.7%. The tumor cells in the present case stained CK7 (+), CK20 (−), and CD10 (−) [5].

Treatment preference in the past has included a range of local excisions to more aggressive treatment composed of anterior exenteration, chemotherapy, or radiation. Local excision more frequently resulted in local recurrence or distant metastasis than did aggressive treatment. In 73% of patients treated with local excision only, treatment failed, with either local recurrence or distant metastasis [6]. When only radiation treatment was administered, disease-free survival was about 33%. Anterior exenteration with urinary diversion showed more promising results, with 87% of patients being disease-free at 6 months to 2 years [7].

Laparoscopic radical cystectomy has recently become the accepted gold standard treatment modality for patients with muscle-invasive bladder cancer [8]. With the development of laparoscopic equipment, laparoscopic radical cystectomy with urinary diversion has become feasible and provides many intraoperative and postoperative advantages over the traditional open approach [9]. A review of the literature found only one reported case of the laparoscopic approach in patients with a urethral tumor [9]. However, laparoscopic surgery in this setting of adenocarcinoma arising from the urethral diverticulum has not previously been reported in the English literature. Although the oncologic outcome of the laparoscopic approach is still being evaluated, many reports have shown that oncologic safety and functional outcome of laparoscopic cystectomy are similar to those of the traditional open approach. Castillo et al first reported on laparoscopic radical surgery in male patients with urethral cancer [10]. He performed a laparoscopic radical cystoprostatectomy and en bloc urethrectomy by additional perineal incision, and this maneuver was a feasible method for the treatment of urethral cancer in men [10]. However, in the current case, the female urethral cancer did not require an additional incision.

There are several key findings in this case. Urethral adenocarcinoma arising from the urethral diverticulum is a rare disease entity. Laparoscopic cystourethrectomy with resection of the anterior vagina in women does not require additional incisions and is a feasible treatment modality for urethral cancer in female patients.

Conflicts of Interest

The authors have nothing to disclose.

REFERENCES

1. Ghoniem G, Khater U, Hairston J, Ramsey A, Woodhouse S. Urinary retention caused by adenocarcinoma arising in recurrent urethral diverticulum. Int Urogynecol J Pelvic Floor Dysfunct 2004;15:363-5.
2. Davis R, Peterson AC, Lance R. Clear cell adenocarcinoma in a female urethral diverticulum. Urology 2003;61:644.
3. Hamilton JD, Leach WB. Adenocarcinoma arising in a diverticulum of the female urethra. AMA Arch Pathol 1951;51:90-8.
4. Takeuchi M, Matsuzaki K, Nishitani H. Clear cell adenocarcinoma of the female urethra: magnetic resonance imaging. J Comput Assist Tomogr 2009;33:142-4.
5. Park SY, Kim BH, Kim JH, Lee S, Kang GH. Panels of immunohistochemical markers help determine primary sites of metastatic adenocarcinoma. Arch Pathol Lab Med 2007;131:1561-7.
6. Choi HG, Son BW, Chung SK, Kim BW. Two cases of primary car-

cinoma of female urethra. Korean J Urol 1994;35:443-7.
7. Awakura Y, Nonomura M, Itoh N, Maeno A, Fukuyama T. Adenocarcinoma of the female urethral diverticulum treated by multimodality therapy. Int J Urol 2003;10:281-3.
8. Cathelineau X, Jaffe J. Laparoscopic radical cystectomy with urinary diversion: what is the optimal technique? Curr Opin Urol 2007;17:93-7.
9. Abascal Junquera JM, Cecchini Rosell L, Martos Calvo R, Salvador Lacambra C, Celma Domenech A, De Torres I, et al. Presentation of a new case of primary clear cell adenocarcinoma of the urethra and its surgical management. Actas Urol Esp 2007;31:411-6.
10. Castillo OA, Pinto I, Portalier P, Diaz MA, Vitagliano G. Laparoscopic radical cystoprostatectomy and in block urethrectomy in urethral cancer: initial experience in 2 cases. Surg Laparosc Endosc Percutan Tech 2007;17:38-41.