A Case of Adenocarcinoma Arising within a Urethral Diverticulum Diagnosed Only by the Surgical Specimen

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Key Words
Adenocarcinoma
Urethral diverticulum
Vaginal cancer

Abstract
We report an extremely rare case of a 50-year-old woman with an adenocarcinoma arising within a urethral diverticulum. She could not be diagnosed correctly before operation and the microscopic pathological examination of the surgical specimen was the only indication showing the cancer origin.

Introduction
Urethral diverticula are not uncommon, with incidence rates of 1-6% [1], and are generally caused by recurrent infections or trauma to the paraurethral glands. However, a carcinoma arising within a urethral diverticulum is extremely rare. The first case was documented in 1951 by Hamilton and Leach, and there are 68 cases in the literature to date [2]. Described here is a case of adenocarcinoma arising within a urethral diverticulum which could not be diagnosed correctly before operation.

Case Report
A 50-year-old woman, gravida 2, para 2, visited a urologist because of urinary incontinence 2 months ago. Since urethral endos-copy and the urine examination pointed out no malignancy in her urinary tract, she was referred to a gynecologist. She was pointed out to have a tumor in the anterior vaginal wall and received a punch biopsy which revealed a vaginal adenocarcinoma. Then, she was referred to our hospital. Since primary adenocarcinoma in the anterior vaginal wall is very rare [3], she was instructed to have the following examinations: cervical Papanicolaou’s smear test, gastrointestinal tract screening, urinary tract screening, breast screening, chest X-ray, computed tomography, magnetic resonance imaging (MRI) and abdominal ultrasonography. Urethral endoscopy showed no abnormal urethral lesion and no abnormal cell was detected in her urine by cytological examination. There was also no abnormal finding in the uterus, the gastrointestinal tract, the breast and the lung. The MRI showed that the tumor was 3.5 × 1.0 × 1.0 cm in size and located between the urethra and
the vagina, and suspected to be of urethral origin (fig. 1). An aspiration smear test through the vaginal wall and a biopsy through the urethral wall indicated a mucin-producing adenocarcinoma. Pathological findings indicated that surgical resection seemed better than radiation and chemotherapy. There was no evidence of distal malignancy and the tumor could be resected surgically. She, therefore, received an anterior pelvic exenteration, with pelvic lymphadenectomy and ileal conduit. Macroscopic examination of the surgical specimen showed a triangle-shaped crater measuring 3 cm in length in the anterior vaginal wall, but no malignant finding in the posterior urethral wall (fig. 2). The origin of the cancer was still unclear at the time of surgery, but microscopic pathological examination showed that the tumor was filled with mucin, and originated from a small urethral diverticulum (fig. 3). The surface of the urethral wall was almost intact but several skip lesions of tumor invasion were located between the urethral diverticulum and the anterior vaginal wall. The patient was followed for 10 months, and remains without recurrent tumor.

Fig. 1. Pelvic MRI (sagittal image by dynamic MRI). Arrowheads indicate the tumor.
Fig. 2. Macroscopic findings of the surgical specimen. Left: a triangle-shaped crater lesion in the anterior vaginal wall (arrowheads). Right: posterior urethral wall (arrows) which shows no abnormal finding.
Fig. 3. Photomicrograph of urethral diverticulum. Hematoxylin and eosin stain. × 160.

Conclusion
An extremely rare case is reported of a 50-year-old woman with an adenocarcinoma arising within a urethral diverticulum. She could not be diagnosed correctly before operation and the microscopic pathological examination of the surgical specimen was the only indication showing the cancer origin. We gynecologists should consider a carcinoma of the urethra in any case of undiagnosed bladder outlet obstruction. This case, however, showed no abnormal finding in the urinary tract.

References