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Nephrogenic adenoma arising in a complex circumferential urethral diverticulum of a female patient with a history of childhood neuroblastoma and chemotherapy

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A 32-year-old African American woman presented with worsening frequency, urgency, and incomplete bladder emptying. Her past medical history was significant for a paraspinal neuroblastoma at the age of 4 years, which was treated by surgical resection, Adriamycin®, and cyclophosphamide. Physical examination revealed a large mass in the anterior wall of the vagina. Magnetic resonance imaging and cystoscopic evaluation revealed an ostia and bulging mass extending from the bladder neck to beyond the external sphincter elevating the bladder neck, as seen in male patients with a prominent median lobe of the prostate (Figs. 1, 2). On urodynamic testing she had a maximum cystometric capacity of 126 mL and a maximum flow rate of 5.6 mL/s, indicating bladder outlet obstruction. Secondary to her refractory voiding symptoms and the possibility of malignancy, surgical excision was pursued. The urethral diverticulum was excised transvaginally through an “Inverted U” incision. Resection was evaluated using intraoperative ultrasound and frozen sections showed no atypia or malignancy. The urethra was reconstructed in two layers and a Martius fibrofatty flap was used to fill the defect site. Postoperatively, the patient did not demonstrate any stress urinary incontinence and her voiding symptoms improved, with a maximum flow of 30 mL/s. Final pathology revealed nephrogenic adenoma (Fig. 3).

Nephrogenic adenoma (NA) is a rare metaplasia that most often arises in the urinary bladder; however, 15 % may arise in the urethra, and it is predominantly diagnosed in male

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Compliance with ethical standards

Conflicts of interest

None.

Consent

Written informed consent was obtained from the patient for publication of this Images in Urogynecology article and any accompanying images.

patients. Recurrent infection, anatomical abnormalities, and a history of urinary tract instrumentation predispose patients to the development of nephrogenic adenomas [1, 2]. Unique to this case is the patient's history of cyclophosphamide administration, which is a known risk factor for bladder cancer [3]. Bladder malignancy may occur a few decades after exposure; thus, lifelong surveillance may be warranted. In addition, as nephrogenic adenoma and urethral diverticuli tend to recur, ongoing surveillance with cystoscopy and pelvic MRI is recommended.

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Fig. 1.

Magnetic resonance imaging revealed a multiseptated/multilocular cystic mass (*arrows*) circumferentially encasing the entire length of the urethra, anterior to the vagina, and reaching the base of the bladder, measuring $3.8 \times 2.7 \times 3.7$ cm. No definite solid components are identified. The findings are suggestive of congenital versus complicated diverticulum and are unlikely to be malignant

Fig. 2.

Left: cystoscopic view of the distal urethra. The external urethral sphincter is seen proximally (*white asterisk*) and the roof of the diverticulum is seen distally (*arrows*). *Right:* looking cystoscopically through the neck of the diverticulum. The opening to the diverticulum is seen (*arrow*) and the thin roof covering the rest of the urethral diverticulum is also appreciated (*black asterisks*)

Fig. 3. Nephrogenic adenoma (H&E, $\times 200$). High power image shows tubules (*arrows*) and microcysts (*asterisks*) lined with a single layer of bland cuboidal cells with clear cytoplasm. There are prominent inflammatory infiltrates, dilated vessels, stromal edema, and myxoid changes