

Leiomyoma in a female urethral diverticulum

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Abstract Neoplasms arising within urethral diverticula are rare. It is important to know if a diverticulum is filled by tumor, but traditional diagnostic methods, such as voiding cystourethrography, cannot detect a tumor or diverticulum, as in our case. We report an unusual case of leiomyoma developing in a female urethra diverticulum and review the literature.

Key words Leiomyoma · Urethral diverticulum · Female · CT · MRI

Introduction

Urethral diverticulum in women is a rare entity, with an incidence ranging from 0.6% to 6.0%.¹ The occurrence of a tumor in a urethral diverticulum is rare,^{2,3} although benign, premalignant, and malignant tumors have been detected there. This article reports an unusual case of a leiomyoma in a female urethral diverticulum.

Case report

A 49-year-old woman presented with a complaint of painless macroscopic hematuria of 7 years' duration. She also reported that she had noticed, by palpation, the presence of a painless mass along the anterior vaginal wall. Physical examination revealed a nontender mass in the anterior wall of the vagina.

Her urine culture was negative, and routine laboratory investigations were normal. At cystourethroscopy and voiding cystourethrography (VCUG), there was no evidence of a urethral diverticulum. Transvaginal ultrasonography (US) at that time showed a 3.5 × 2.5 cm solid mass located along the anterior aspect of the vagina but not attached to the vagina (Fig. 1). Computed tomography (CT) and magnetic resonance imaging (MRI) demonstrated a solitary, solid, well-defined mass along the urethra (Figs. 2, 3). On MRI, the mass was isointense to muscle on T1-weighted images and slightly hyperintense on T2-weighted images. After intravenous gadolinium administration, the mass showed diffuse and homogeneous enhancement.

The mass was excised under spinal anesthesia, and the urethral defect was reconstructed with a 4-0 absorbable suture. She had an uneventful postoperative course.

Histopathological evaluation demonstrated a leiomyoma arising in a urethral diverticulum. Examination of sections revealed that the lesion consisted of cells with a blunt nucleus and eosinophilic cytoplasm that formed bundles. Areas of focal hyalinization were observed. There was no marked mitosis or cytological atypia. Diffuse staining with desmin and actin was observed in the immunohistochemical (IHC) examination (Fig. 4).

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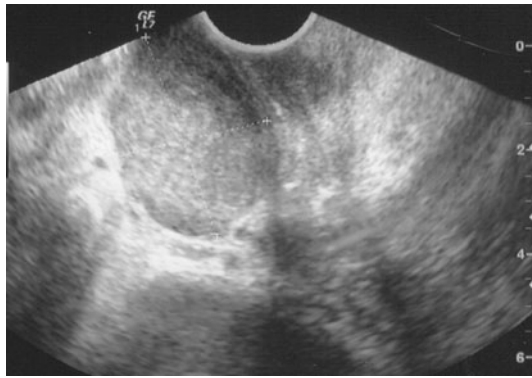


Fig. 1. Transvaginal sonogram. The diverticulum is filled with the tumor mass



Fig. 2. Computed tomography reveals a periurethral tumor (arrow)

Fig. 3. Axial T1-weighted (a), axial STIR (b), sagittal T2-weighted (c), and postcontrast sagittal T1-weighted (d) images demonstrate a large T1 isointense, T2 slightly hyperintense, markedly enhanced, well-defined mass (arrows) lying along the urethra

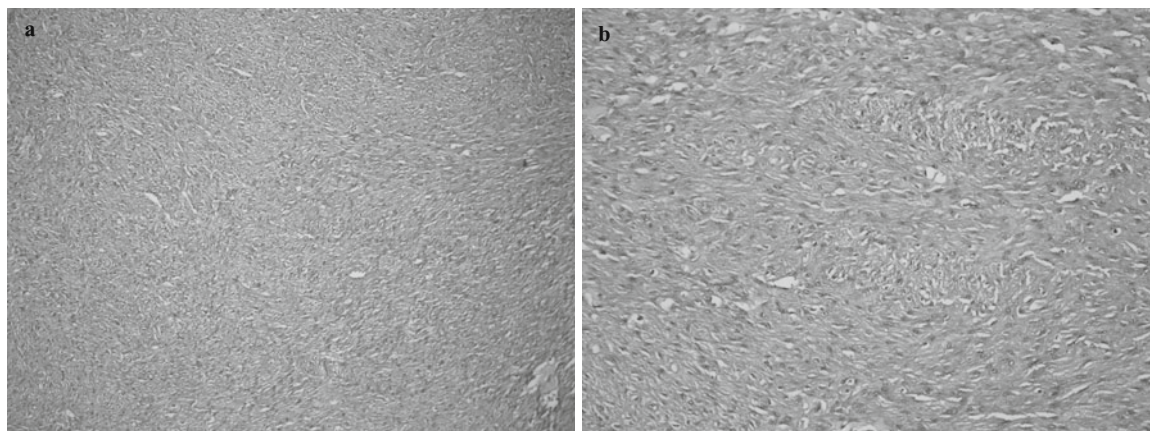
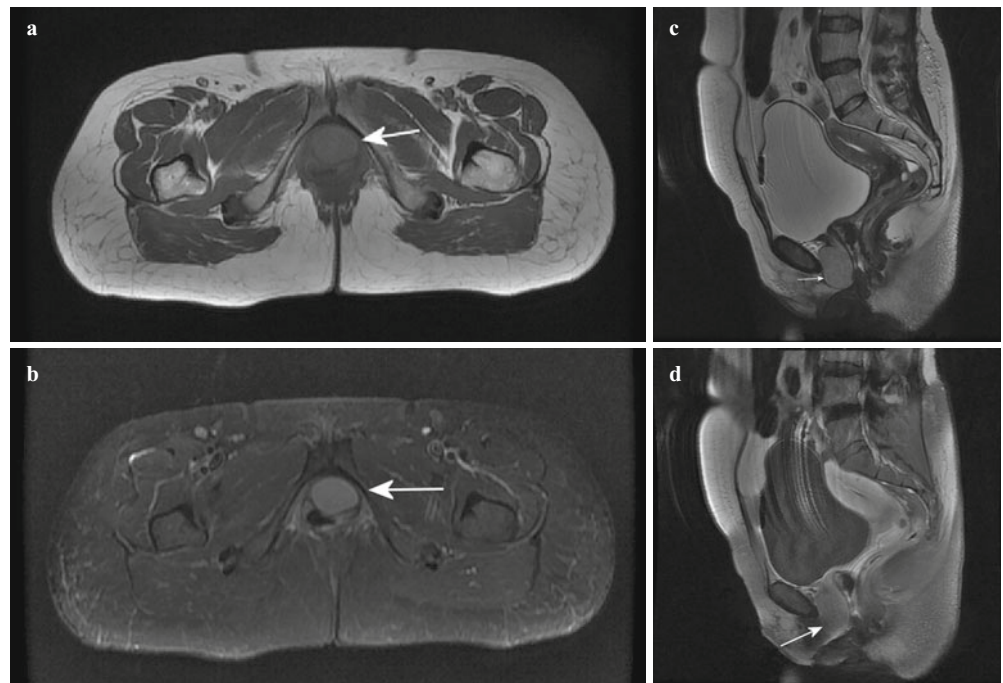


Fig. 4. a Histology of the operative specimen shows spindle cells arranged in bundles (H&E, $\times 100$). b High-power view of the spindle cells showing no significant atypia or mitotic figures

Discussion

Urethral diverticula may be congenital or acquired. Whereas most urethral diverticula are congenital in males, those in females are usually acquired. The etiology is uncertain, although it has been suggested that acquired diverticula may develop as a result of rupture of dilated and infected periurethral glands.^{1,4} In addition, urethral injury during childbirth and urethral or vaginal surgery may cause a urethral diverticulum. The incidence of urethral diverticulum is lower in white women (1.1%) than in black women (6.0%),³ who account for 78%–86% of female urethral diverticulum cases.⁴ Urethral diverticula are usually located in the posterolateral wall of the middle third of the urethra.¹ Calculi may occur in 1.5%–10.0% of female urethral diverticula.⁵ Approximately 25% of the lesions are multiloculated.⁶

The symptoms of the urethral diverticula are not specific, although patients may complain of dysuria, polakiuria, dyspareunia, urinary incontinence, recurrent urinary tract infection, and dribbling after voiding.^{1,6}

Voiding cystourethrography has traditionally been used for the initial diagnosis of urethral diverticulum. However, VCUg has a false-negative rate as high as 35% for detecting a urethral diverticulum.⁷ Although urethroscopy has an overall diagnostic accuracy of 90%, it does not enable determination of the real size of a diverticulum or evaluation of the diverticular wall. Recently, US, CT, and MRI have been successfully used for comprehensively evaluating urethral diverticula.

A neoplasm arising in a urethral diverticulum is rare. Urethral diverticular carcinomas, endometriosis, and nephrogenic adenomas have been reported in the English-language medical literature.^{8,9} Chronic inflammation, stasis, and mucosal irritation may lead to stone formation or metaplastic or neoplastic changes in the urethral diverticulum. The first report of carcinoma in a female urethral diverticulum appeared in 1951, authored by Hamilton and Leach.¹⁰ Carcinoma arising in a urethral diverticulum accounts for 5% of all urethral carcinomas.² Squamous cell carcinoma is the most common type of urethral carcinoma, whereas 40%–60% of carcinomas arising in a urethral diverticulum are adenocarcinomas.^{7,9} Urethral diverticular malignancies characteristically present late with early metastases, so there is a high risk of recurrence after surgery. Symptoms are often nonspecific and related to irritation of the lower urinary tract.

Leiomyoma of the female urethra is a rare benign tumor derived from smooth muscle tissue. Leiomyomas typically show low to intermediate signal intensity on T2-weighted MRI images. To the best of our knowledge, review of the English literature reveals no cases of leiomyoma arising from a urethral diverticulum.

Conclusion

Sometimes a urethral diverticulum is filled with tumor and cannot be visualized at cystourethroscopy or VCUg, as was seen in our case. Clinicians should be aware of the possibility of a urethral diverticulum when patients have lower urinary tract irritability symptoms even if cystourethroscopy and VCUg are normal. As in our case, cross-sectional imaging tools can improve diagnostic accuracy in a patient presenting with the complaint of lower urinary tract irritability symptoms.

References

- Hosseinzadeh K, Furlan A, Torabi M. Pre- and postoperative evaluation of urethral diverticulum. *AJR Am J Roentgenol* 2008;190:165–72.
- Rajan N, Tucci P, Mallouh C, Choudhury M. Carcinoma in female urethral diverticulum: case reports and review of management. *J Urol* 1993;150:1911–4.
- Gonzalez MO, Harrison ML, Boileau MA. Carcinoma in diverticulum of female urethra. *Urology* 1985;26:328–32.
- Tines SC, Bigongiari LR, Weigel JW. Carcinoma in diverticulum of the female urethra. *AJR Am J Roentgenol* 1982;138:582–5.
- Josef B, Raeto TS. Giant calculi in urethral diverticula. *Can Med Assoc J* 2008;178:994.
- Guidi HG, Montelatto NI, Ribeiro RM, Pinotti JA. The treatment of female urethral diverticulum with calculus through ultrasonic lithotripsy. *Int J Gynaecol Obstet* 1993;41:277–81.
- Siegel CL, Middleton WD, Teefey SA, Wainstein MA, McDougall EM, Klutke CG. Sonography of the female urethra. *AJR Am J Roentgenol* 1998;170:1269–74.
- Summitt RL Jr, Murrmann SG, Flax SD. Nephrogenic adenoma in a urethral diverticulum: a case report. *J Reprod Med* 1994;39:473–6.
- Thomas RB, Maguire B. Adenocarcinoma in a female urethral diverticulum. *Aust N Z J Surg* 1991;61:869–71.
- Hamilton JD, Leach WB. Adenocarcinoma arising in a diverticulum of the female urethra. *AMA Arch Pathol* 1951;51:90–8.