Case Report

Unusual Prolapse of Anterior Urethral Wall Presenting as A Vulval Mass in A Woman

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INTRODUCTION

The adult female urethra is about 4 cm long and extends from the bladder neck to the vaginal vestibule. Its lining changes gradually from transitional to nonkeratinized stratified squamous epithelium. Instead, the vaginal vestibule lining is keratinized stratified squamous epithelium. Urethral prolapse usually denotes the complete circular eversion of the urethral mucosa through the external meatus. Two different entities exist: premenarcheal and menopausal urethral prolapse. Premenarcheal prolapse is predominantly asymptomatic and is usually brought to medical attention by vaginal bleeding. This kind of urethral prolapse can be diagnosed by its typical clinical appearance. The menopausal group seeks medical attention primary because of the severity of urinary symptoms, ie, nocturia, urgency, tenesmus, dysuria, and frequency. Normally, the diagnosis of urethral prolapse is made by verifying that a central opening is present within the prolapsed tissue and that this opening is the urethral meatus. We herein report an unusual type of urethral prolapse limited to its anterior wall in a 73-year-old female.

CASE REPORT

A 73-year-old woman, gravida 6 para 6, presented to our OPD complaining of a mass protruding from the vulva and blood-tinged toilet paper after wiping after voiding for several weeks. She had no frequency, urgency or other lower urinary tract symptoms except mild stress urinary incontinence during coughing. On physical examination, an erythematous tumor-like lesion with smooth surface, about 2.3 cm in diameter, was noted on the vestibule at the original site of the external urethral orifice. Urethral catheterization helped identification of the urethral meatus, and the real external urethral orifice was found underneath the mass. There was no anterior vaginal wall prolapse, nor cystocele under Valsalva maneuver. Excretory urogram showed normal bladder and kidneys and minimal postvoid residual urine. Cystourethroscopy revealed prolapse of the anterior urethral wall, but the posterior lip of the external urethral orifice was normal. Mild downward angulation of the distal urethra was also noted. There was no urethral diverticulum, bladder diverticulum, or other intravesical lesions. After placing a Foley catheter into the urethra as a guide, the patient was treated surgically. Partial wedge resection of the skin and soft tissues overlying the mass was done first for pathologic studies. There were no serous or purulent secretions, nor cystic lesion noted in the soft tissues overlying the distal urethra. Then, the anterior urethral wall was manually reduced back and was secured in its normal position through sequential multi-layered plications of the soft tissues around the long axis of the urethra to avoid shortening of the...
total urethral length. Cystourethroscopy was performed immediately after surgical correction and revealed restoration of the normal urethral lumen without residual angulation of the distal urethra. Finally, a Foley catheter was left in place to allow continuous bladder drainage. The indwelling catheter was removed 2 days postoperatively. Histopathological study of the wedge resected specimen showed the lining epithelium was keratinized stratified squamous epithelium; the subcutaneous tissue showed edematous change with only scanty inflammatory cells infiltration; no tumor cells were found. Follow-up bladder scan was performed 2 weeks postoperatively which revealed less than 15 c.c of postvoid residual urine. After five months of postoperative follow-up the vulva looked normal and the patient had no more complaint of vulval discomfort, recurrent vulval mass or stress urinary incontinence.

**DISCUSSION**

The female urethra is composed of inner longitudinal and outer circular-oblique smooth muscle layers. Usually, a natural cleavage plane is present between the inner and outer muscle layers. In a healthy female urethra, this cleavage plane firmly adheres to the opposing muscle layers. Separation of these muscle layers may be induced by recurrent episodes of increased intra-abdominal pressure, such as heavy coughing, constipation, or obesity. Poor attachments between the longitudinal and circular, oblique, smooth muscle layers and the mucosa of the urethra results in complete and circular eversion of the urethral mucosa through the external meatus and leads to urethral prolapse. Other contributing factors such as trauma, malnutrition, urinary and vaginal infections, and urethral mucosa redundancy have been postulated in the etiology of this condition. Urethral catheterization or cystourethroscopy help to verify the presence of the urethral meatus in the center of the prolapsed mass. Routine evaluation with intravenous pyelogram and voiding cystourethrogram is unnecessary except in cases in which there is doubt concerning malignancy, prolapsed ectopic ureterocele, or abnormalities of the ureterovesical junction. Common presenting complaints included vaginal bleeding, vaginal discharge and mass protruding from the urethral opening.

The differential diagnosis of urethral prolapse should include urethral malignancy, urethral caruncle, ectopic ureterocele, condyloma, and rhabdomyosarcoma. Urethral masses that were actually sarcoma botryoides or endodermal sinus tumors have been misdiagnosed as urethral prolapse. Urethral leiomyomas and malakoplakia have also been misdiagnosed as urethral prolapse.
Our unusual case was due to prolapse of the anterior wall of the urethra with the external urethral orifice located not in the center but underneath the prolapsed mass. It is quite different from most of the reported cases in the literature, which involves prolapse of the circumferential mucosa of the distal urethra rather than the anterior urethral wall only. Incisional biopsy of the vulval mass in our case demonstrated that the lining epithelium was keratinized stratified squamous epithelium, which is different from the urethral mucosal lining but is consistent with the vulvar vestibular epithelial lining. Gross appearance and cystourethroscopy findings clearly demonstrated that the anterior urethral wall was prolapsed, which further pushed the overlying vaginal vestibule out to form a vulval mass.

The exact cause of urethral prolapse in our case is not known. There was no trauma, chronic cough, or chronic constipation histories in this case. Also, pelvic floor weakness due to repeated vaginal deliveries or menopause cannot be attributed to the contribution factors of the urethral prolapse, since our case had no cystocele or severe stress urinary incontinence. The mechanism of this unusual anterior urethral wall prolapse could be due to intrinsic weakness of the vestibule of the vulva above the external urethral meatus which caused progressive protrusion of the poorly fixed anterior wall of the distal urethra, although this assumption needs to be further clarified.

For women with mild forms of urethral prolapse, the recommended therapy consists of local hygiene with sitz baths and topical antibiotic, steroid, or estrogen creams.9 If medical therapy does not rapidly reduce the prolapse, surgery is the treatment of choice.10 Since the urethral prolapse encountered in our case was not limited to the mucosa but involved the full thickness of the anterior urethral wall and the patient also had mild stress urinary incontinence, excision of the protruded mass was not justified because this would result in even shorter of the whole urethral length. Our surgical procedures described above would not jeopardize the original length of the urethra. The mechanism of mild stress urinary incontinence noticed preoperatively could be due to poor fixation of the distal urethral wall when the prolapse occurred which decreased the functional urethral length. After surgical fixation of the distal urethral wall and restoration of the normal anatomic position, the stress urinary incontinence disappeared even after five months of postoperative follow-up.

REFERENCES