Leiomyoma of the female urethra: a case report and review of the literature

Leiomioma de uretra femenina, aportación de un caso clínico y revisión de la literatura

Dear Editor,

We present the case of a 23-year-old female with unremarkable history who consulted with the urology service for dysuria and repeat urinary infections (cystitis) that did not respond to antibiotic therapy. She also complained of a 4-months old excrescent lesion in the urethral meatus which was painless, firm, and had been increasing in size in the past 4 weeks.

The physical examination revealed an exophytic, whitish-pink, rounded, firm lesion with well defined borders, not adhered to deep tissues, and joined to the distal urethra, but not protruding into the meatus.

Due to the clinical suspicion of leiomyoma of the urethra, surgery was scheduled.

The lesion observed during the operation was whitish, oval, 1.5-cm in diameter, pedunculated, and located in the distal urethra. A simple excision of the lesion was done.

The anatomopathological examination revealed a well-delimited lesion consisting of bundles of fusiform cells, with no mitoses, no atypia, and good cell differentiation. The immunohistochemical test was positive for actin.

Thus, the histopathological diagnosis of leiomyoma of the urethra was confirmed.

Uterine leiomyoma is the most common tumor in women; leiomyoma of the urethra, however, is a rare condition[1-10].

The tumor was first described by Buttner in 1984. Bartolozzi et al[10] found 110 cases of these tumors in the literature in English.

The tumor often appears during the woman's fertile age (from menarche to menopause); the mean age of the appearance is around 41 years[2,3].

Its origin is yet unknown[2,3]. It is known that ovarian hormones favor its development, but it is difficult to say how exactly these hormones affect the development of the tumor, as there are documented cases in postmenopausal women[9].

Ovarian hormones act through specific receptors located in the cell nucleus. Receptors are found in the tissues of reproductive organs, lower urinary tract, and cardiovascular system, but other tissues of the body that are affected by these hormones do not express these receptors.

In fact, hormone therapy is accepted by all authors as a symptomatic treatment for leiomyoma[1-10].

Progestogens, rather than estrogens, seem to be the main cause of this tumor, as suggested by the findings in leiomyoma biopsies during the course of the disease. Kesari et al published the first report documenting the existence of estrogen receptors. In most cases, immunohistochemical tests reveal ovarian hormone receptors of the ERα, ERβ and PR types in the nuclei of the tumor cells. These findings, along with the fact that increased levels of ovarian hormones cause an increased ratio of tissue mitosis in leiomyomas, support the theory that these hormones may promote tumoral growth[9].

Leiomyomas can arise in any location in the urinary tract, but the kidney and the bladder (in this order) are the most common sites[4]. In the urethra, the most commonly affected area is the proximal portion; the mean diameter is 3.7 cm, according to the data collected by Bartolozzi et al[10].

The tumors can be asymptomatic or be part of an acute urine retention picture[1-10], and cause symptoms such as dyspareunia, hematuria, space occupying lesion in the urethra, urinary tract infection, and irritative symptoms (the latter two are the most frequent, according to Fry et al and Moopan et al)[5,6].

Diagnosis is based on the clinical history, physical examination (a mass in the urethral lumen, visible if the leiomyoma is located in the distal urethra), and imaging techniques; in any case, the final diagnosis is given by histopathological examination.

Contrast radiological tests such as retrograde cystourethrography and voiding cystography permit to identify repletion defects in these patients. Urethrocystography permits the direct visualization and identification of these lesions, and sampling for biopsy.

The ultrasound shows hypoechogenic, well-defined, and homogeneous lesions[7].

The study published by Pavlica[9] highlights the importance of imaging techniques in this condition and the fact that transvaginal ultrasound and MRI can identify the benign nature of the lesion.

Patients with this disease often present with repeat urinary tract infections due to the obstruction to the flow of urine and the stagnation of urine inside the urinary tract. The study by Goto et al used urodynamic testing to corroborate the existence of this flow obstruction. Patients with leiomyoma of the urethra have high detrusor pressure and low maximum flow, with significant post-void residual volume in voluntary voiding, and elevated urethral resistance. Urodynamic testing after surgical correction shows a normalization of the detrusor pressure at maximum flow, and no post-voiding residue; this translates into an improvement of the urination difficulties and dyspareunia.

Differential diagnosis of this condition should include: urethral caruncle, extravesical bladder leiomyoma, plasmacytoma, and lymphoma; the lesion must be differentiated from leiomyosarcoma (characterized by cellular

atypia, pleomorphism, and a high rate of mitosis found upon pathological examination of the lesion).

The most appropriate treatment for this condition is local excision[1-10]; the urethral mucosa could be injured during surgery, and must be repaired (due to the possible development of urethral stenosis or stress incontinence as complications); if the leiomyoma is removed with no disruption of the urethral mucosa, it most likely does not originate in the smooth circular muscle of the urethra.

There usually is no recurrence or malignization after treatment; however recurrence was reported in one case-a patient with a tumor of more than 8 cm in diameter.

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