



FEMALE PARAURETHRAL LEIOMYOMA: A RARE ENTITY

Urology

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ABSTRACT

Leiomyomas are rare benign fibromuscular tumors that originate from smooth muscle cells. Paraurethral leiomyoma is a rare, benign hormone dependent tumor of mesenchymal origin affecting women of reproductive age group. It consists of 5% of all paraurethral masses occurring in 1:1000 women. They arise from smooth muscles of urethra and usually present with symptoms of pelvic pressure, obstructive and storage lower urinary tract symptoms, and a protruding mass. Herein, we report a case of 30year old woman presented with palpable paraurethral mass and predominantly obstructive Lower Urinary Tract Symptoms. She underwent clinical examination, USG and MRI imaging followed by Cystourethroscopy and Surgical excision of the mass. Histopathological examination confirmed paraurethral leiomyoma.

KEYWORDS

Leiomyoma, paraurethral, benign, mass, excision

INTRODUCTION

Leiomyomas are rare benign fibromuscular tumors that originate from smooth muscle cells.¹ Paraurethral leiomyoma is a rare, benign hormone dependent tumor of mesenchymal origin affecting women of reproductive age group.² It consists of 5% of all paraurethral masses occurring in 1:1000 women.³ Only few cases of true paraurethral tumors have been reported amongst the reported cases of urethral neoplasms.³ They arise from smooth muscles of urethra and usually present with symptoms of pelvic pressure, obstructive and storage lower urinary tract symptoms, and a protruding mass.⁴ Fifty percent of the patients are asymptomatic.² The rarity of this condition together with an overlap of its clinical manifestations with other common urogenital and gynaecological conditions often leads to a misdiagnosis until histopathology is performed. It is also essential to preoperatively distinguish urethral or paraurethral origin of the mass, to effectively plan an appropriate surgical approach and avoid undue complications. Herein, we report a case in a 30-year-old female patient who presented with a mass at paraurethral region which turned out to be leiomyoma on histology.

Case Capsule

A 30year old female attended the OPD with chief complaints of a lump in her urethra in the last six months. The patient accidentally felt the lump while wiping and cleaning her genital. She had symptoms frequency and urgency of urine, poor flow, straining while passing urine and sensation of incomplete emptying of urine. There was no associated menstrual complaint or bleeding from the lump or any history of pain in abdomen. She also denied history of other lower urinary tract symptoms complaints, dysuria, burning micturition, hematuria, cloudy urine, lithuria and dyspareunia.

Past medical and surgical history was insignificant. General examination was normal. On physical examination, the abdomen was soft, non-tender, no organomegaly or lump was elicited. On local examination of the perineum revealed a single well defined, non-tender, firm, ovoid swelling measuring 2x3.5cm in the left anterior aspect of urethra from 12'o clock to 6'o clock position. Blood investigations, urinalysis and urine cytology were within the normal ranges

Further, radiological investigations were performed. Ultrasonography of whole abdomen and pelvis revealed a well-circumscribed heterogeneously hypochoic lesion along the left anterolateral aspect of the urethra, measuring 29 x 28 x 24mm. Internal vascularity was noted on colour doppler study. The lesion was seen to cause mass effect and effacement of the urethra. Urinary Bladder showed circumferential wall thickening with multiple trabeculations,

maximum wall thickness measured 8mm. Her Pre-void volume was 730 ml and Post-void residue was 464 ml, which was significant. Above findings were suggestive of chronic bladder outlet obstruction changes. Uterus and Ovaries appeared normal. Patient was then subjected to MRI Pelvis. It showed a solitary, well-circumscribed eccentric solid mass lesion measuring 3.0(AP) x 2.5(W) x 2.5(H) cm in a retropubic-subpubic location, abutting the posterior cortex of pubic bone and indenting the bladder base. On axial cuts, the lesion is epicentered along the anterior and left lateral wall of the urethra, between the 12 and 6'o clock positions, and severely narrowing the urethral lumen. Lesion has differential signal on the T2-W images comprising of a hypointense core surrounded by an intermediate signal rim, with the core hypo-enhancing relative to the outer rind on the post contrast images (Figure 1). The lesion appeared to indent the anterior wall of the lower third of the vagina without invasion. Fat planes in the left paraurethral region were preserved. No bony erosion noted. Urinary bladder was distended with thick and trabeculated walls. Perivesical fat planes were normal.

After consenting and comprehensive discussion, patient was scheduled was Rigid Cystourethroscopy with Excision of Left Paraurethral mass under Spinal Anaesthesia. Cystourethroscopy findings revealed- Urethra was normal, Bladder was grossly trabeculated (Changes of Bladder Outlet Obstruction) (Figure 2). Bilateral Ureteric orifices appeared normal. After urethral catheterization with 16-French Foleys catheter, hydro-dissection was done with injection of normal saline. Longitudinal incision was taken 2cm lateral to labia majora, over the paraurethral mass area. Mass enucleated (Figure 3). Dead space was obliterated with Vicryl 2-0 sutures. Post-operative was uneventful and 16 Fr Foleys catheter was left in place for 10 days (Figure 6).

The excised specimen was well defined, encapsulated, firm, greyish-pink tumor measuring 3.5 x 2.7 x 2.4 cm (Figure 4). Cut section revealed tan with central calcified area, with whitish whorled pattern. Histopathology report showed haphazard and vague fascicular arrangement of spindly tumor cells on 10X magnification, and tumor cells showed indistinct cytoplasm with plump oval nuclei showing tiny nucleoli on 40X magnification (Figure 5). Immunohistochemistry (IHC) for Smooth Muscle Antigen (SMA) showed diffuse positivity. Histopathological and IHC features were consistent with Paraurethral Leiomyoma.

Post-operative clinical course was uncomplicated. The patient was discharged on 4th day with Foleys catheter in situ. Patient was followed-up on regular basis and Foleys catheter was removed on the 10th day. Following the catheter removal, patient did well and voided

with good flow. The patient was evaluated at 1 month and 3 months follow-up session, the previous complaints of Lower Urinary Tract Symptoms were relieved, and the surgical site was clean and healed well. No surgical complication and new onset of Lower Urinary Tract Symptoms or incontinence was observed after surgery.

DISCUSSION

Paraurethral leiomyomas are mesenchymal benign tumors.⁵ The etiology is unknown but majority of the cases occur in reproductive age women, thus explains the hormonal role in its development.⁶ Due to close anatomical relationship and positioning, distinction between vaginal, urethral and paraurethral leiomyoma is often very difficult. In addition, they have similar clinical presentation. Paraurethral leiomyomas have a usually occurs between posterior urethral wall and anterior vaginal wall in the paraurethral space or vesicovaginal septum. It affects both the genders but it is more prevalent in females. The usual age of presentation is around 40-44 years.⁷ Most common symptoms are voiding dysfunction Lower Urinary Tract Symptoms, vaginal bleeding, dyspareunia, symptoms of pelvic pressure, urinary retention or protruding mass.⁴ Paraurethral leiomyomas can also appear as an asymptomatic mass.¹ The differential diagnosis of paraurethral mass can be an urethral mass (leiomyomas), urethral prolapse, urethral diverticulum, urethral caruncle, Bartholin's duct cyst, Bartholin's abscess, Skene's duct abscess, Gartner's duct cyst or urethral carcinoma.³ Malignant lesions including adenocarcinoma, sarcomas and squamous cell carcinoma, are the other most important differential diagnoses that should be ruled out.

Paraurethral leiomyomas are hormonally sensitive. They have numerous estrogen receptors. They grow during pregnancy and regress after menopause.⁸ Periurethral masses are uncommon, with a retrospective case series finding that they comprise less than 5% of patients seen at a tertiary, referral urogynecology practice. Urethral and Paraurethral leiomyoma make up just around 5% of periurethral masses. Urethral diverticulum comprise the majority of periurethral masses and vaginal cysts are a similar percentage of periurethral masses as urethral leiomyoma.⁹

Work-up for Paraurethral masses includes a detailed history and examination, and if the diagnosis is not clear, imaging and cystoscopy may be employed. Radiological imaging techniques like USG, CT scan and pelvic MRI help in exact location and origin of mass and relationship with nearby structures which is essential for surgical planning. Transvaginal or Transperineal USG has a role, which will show well-defined isoechoic or hypoechoic homogeneous mass and inner vascularity on colour flow Doppler. MRI shows leiomyoma as homogeneous well-encapsulated mass appearing hypointense or isointense on T1-weighted images and intermediate signal on T2-weighted images.¹⁰

Local surgical excision via Transvaginal or Transurethral route has been recommended as the treatment of choice.¹¹ For larger paraurethral leiomyomas, Abdominoperineal approach may be necessary. Histopathology is the gold standard for diagnosis which shows interlacing fascicles of spindle-shaped smooth muscle cells with oval nuclei and eosinophilic cytoplasm.¹² Immunohistochemistry is positive for smooth muscle cell antigen, as seen in our case. Given the close relationship of these masses to the bladder and urethra, injury to these organs should be considered as a possible complication of the surgery. Foley catheter insertion helps to prevent urethral trauma and other complications. Clinical follow-up is required for the evaluation of any urinary symptomatology like urinary incontinence. Recurrence is rare. However, stress urinary incontinence post-excision is recognised complication, which is often managed with a mid-urethral sling. Migliari et al. revealed two cases of stress urinary incontinence during the follow-up after paraurethral leiomyoma resection.¹³ This presented case showed no post-surgical complications and the post-operative recovery was uneventful; also, during follow-up periods, she did not face any urinary complications. Symptoms were completely relieved.

It is thus important, that all specialists especially in female pelvic surgical field should be familiar with the broad differential diagnosis and management of paraurethral masses to streamline the patient experience and facilitate favourable patient outcomes.

CONCLUSION

Paraurethral Leiomyomas are rare benign mesenchymal tumors of

female genitourinary tract. They can result in irritative or obstructive Lower Urinary Tract Symptoms, especially in female. The anatomical relationship of the mass with the bladder and urethra poses a challenge for clinicians. Imaging modalities like Ultrasonography or MRI plays an important role in pre-operative diagnosis and surgical planning. Surgical excision of the mass is the treatment of choice and should be performed carefully.

We report a rare case of Paraurethral Leiomyoma in a female presented with Storage and Obstructive Lower Urinary Tract Symptoms where timely surgical excision was performed and symptoms completely resolved post-surgical treatment.

Hence it is very important for the clinicians as well as the pathologists to be familiar with the differential diagnosis of lesions in the urethral and paraurethral region in order to achieve adequate management and favourable patient outcome.

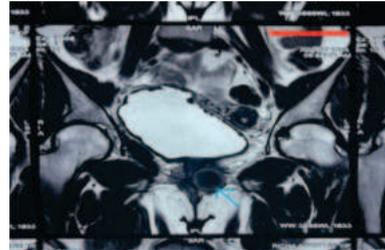


Figure 1- MRI T2WI Axial image showing a well-circumscribed eccentric solid mass lesion measuring 3.0(AP)x 2.5(W)x2.5(H)cm (blue arrow), epicentered along the anterior and left lateral wall of the urethra, between the 12 and 6'o clock positions, and severely narrowing the urethral lumen, comprising of a hypointense core surrounded by an intermediate signal rim, with the core hypoenhancing relative to the outer rind. Urinary bladder appeared distended with thick and trabeculated walls

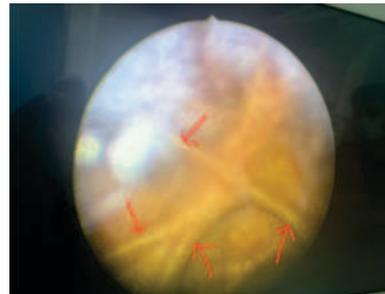


Figure 2- Rigid Cystoscopy showing Bladder Trabeculations (Changes of Bladder Outlet Obstruction) (red arrow)



Figure 3- Intraoperative image of Paraurethral mass being excised



Figure 4- Excised specimen of Paraurethral mass, well-defined, ovoid, encapsulated, greyish-pink measuring 3.5 x 2.7 x 2.4 cm

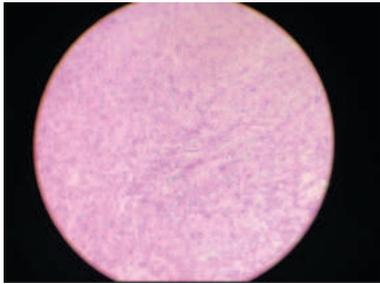


Figure 5- HPE image- Haphazard and vague fascicular arrangement of spindle tumor cells with indistinct cytoplasm and plump oval nuclei showing tiny nucleoli



Figure 6- Post operative image with 16Fr Foleys in situ

Conflicts Of Interest- None

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