Case Report

Unveiling a rare culprit of lower urinary tract symptoms in a reproductive age group female – A case report

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ABSTRACT

Leiomyomas are generally benign lesions originating from smooth muscle cells. However, paraurethral leiomyomas are a rare occurrence with only a few cases being reported in the literature to date. In view of the scarce existing literature regarding paraurethral leiomyoma, their diagnosis and management pose a peculiar challenge. We, hereby, describe the case of a 43-year-old female, who presented with obstructive voiding symptoms – moderate severity for 4 months. A mass of size 2.5 × 2 cm was palpable along the anterior wall of the vagina and the posterior wall of the urethra. On cystoscopy, the urethra appeared compressed. The patient underwent surgical excision of the mass by the transvaginal route which was confirmed to be leiomyoma on histopathology. Paraurethral leiomyomas are infrequent tumors arising from the periurethral smooth muscle and are an important differential diagnosis for paraurethral masses in females of reproductive age group.

Key words: Leiomyoma, Mesenchymal, Paraurethral, Reproductive

Leiomyomas are generally benign lesions originating from smooth muscle cells. They are commonly found in the genitourinary and gastrointestinal systems with uterine leiomyomas being a common encounter [1]. However, paraurethral leiomyomas are a rare occurrence with only a few cases being reported in the literature to date. They originate as mesenchymal tumors from the paraurethral space with no communication with the bladder, urethra, or vagina. They are believed to be hormone-dependent, particularly estrogen, and thus are found in females of the reproductive age group usually around the 4th–5th decade of life. The clinical presentation may vary from asymptomatic individuals to symptoms of dyspareunia, dysuria, lower urinary tract symptoms, or a mass protruding from the vaginal introitus. The diagnosis is often made with a strong suspicion on imaging studies and confirmed on histopathology after surgical excision [1]. In view of the scarce existing literature regarding paraurethral leiomyoma, their diagnosis and management pose a peculiar challenge.

CASE REPORT

A 43-year-old female presented with obstructive voiding symptoms of moderate severity for 4 months. She denied any history of abdominal pain, urinary tract infection, hematuria, or menorrhagia. Past medical and surgical history was not significant.

On clinical examination, the patient was moderately built and well-nourished. There was no pallor, icterus, cyanosis, clubbing, lymphadenopathy, or edema. General physical examination was within normal limits. Per-abdomen was soft and non-tender, with no organomegaly. On local examination, a mass of size 2.5 × 2 cm was palpable along the anterior wall of the vagina and the posterior wall of the urethra. It was painless, firm in consistency, having well-defined margins and a smooth surface.

Routine blood investigations and urine analysis were normal. On transvaginal ultrasound (USG), a well-defined hypoechoic homogenous mass of size 3.2 × 2.9 cm was seen located between the posterior wall of the urethra and the anterior vaginal wall (Fig. 1). Magnetic resonance imaging (MRI) pelvis showed a well-defined lesion measuring 3.2 × 2.8 cm lesion along the posterior wall of the urethra with a mass effect on the anterior vaginal wall. The lesion appeared isointense on T1 (Fig. 2a) and hyperintense on T2 (Fig. 2b). It showed diffusion restriction with a homogenous post-contrast enhancement. Multiple uterine fibroids were also described. The possibility of paraurethral mesenchymal tumor was considered. On cystoscopy, the urethra appeared compressed (Fig. 3). The overlying mucosa was normal. The bladder was unremarkable.

The patient underwent surgical excision of the mass by the transvaginal route. After placing a per urethral catheter, an

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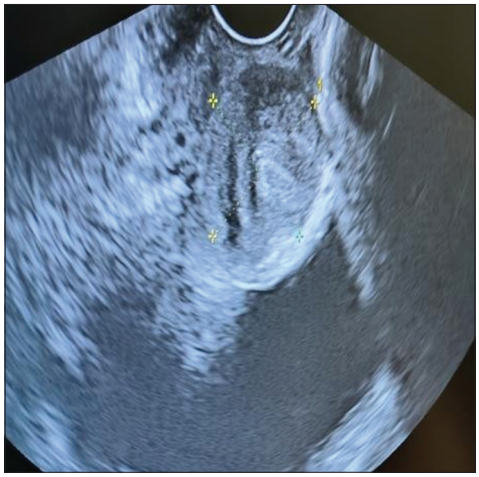

Figure 1: Transvaginal ultrasound shows a well-defined, circumscribed, and hypoechoic homogenous mass of size 3.2 × 2.9 cm in the paraurethral location

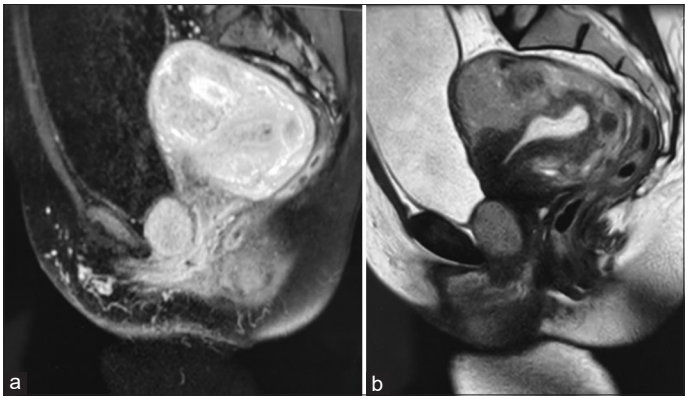

Figure 2: (a) Magnetic resonance imaging image (MRI) (T1) shows a well-defined isointense lesion measuring 3.2 × 2.8 cm lesion along the posterior wall of the urethra with a mass effect on the anterior vaginal wall. The coexisting uterine fibroids can also be appreciated; (b) MRI (T2) with the hyperintense lesion

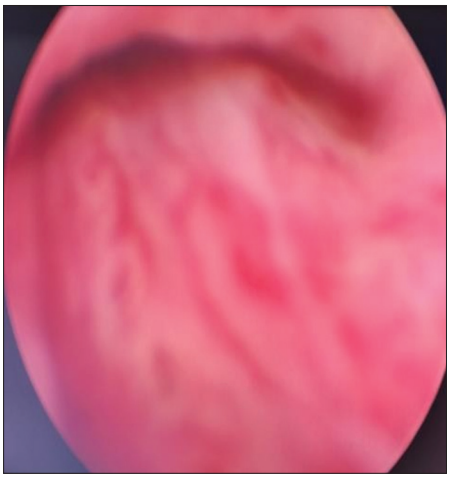

Figure 3: Elevated posterior wall of the urethra on cystourethroscopy

incision was made along the anterior wall of the vagina, and enucleation of the tumor was done after meticulous dissection from the surrounding structures.

Grossly, the tumor was a well-circumscribed mass of size 3.5 × 3 × 1.5 cm with a rubbery consistency (Fig. 4a). Microscopically,

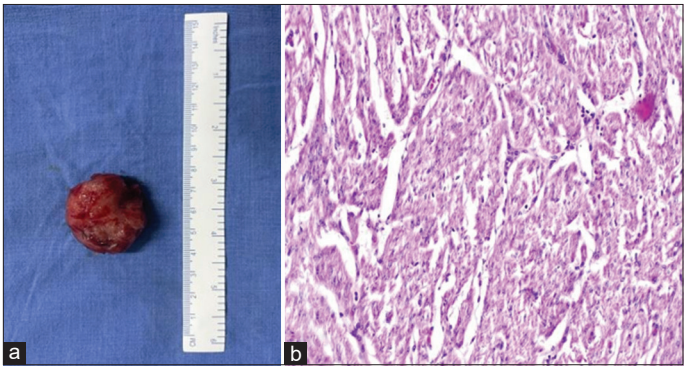

Figure 4: (a) Specimen showing a well-circumscribed mass of size 3.5 × 3 × 1.5 cm; and (b) histopathology image showing smooth muscle cells arranged in the form of interlacing fascicles. The cells have elongated nuclei and eosinophilic cytoplasm under hematoxylin and eosin stain confirming leiomyoma

it was found to have smooth muscle cells arranged in the form of interlacing fascicles. The cells had elongated nuclei and eosinophilic cytoplasm under hematoxylin and eosin stain (Fig. 4b) and hence confirmed to be leiomyoma on histopathology.

The post-operative period was uneventful. The catheter was removed after 1 week. During follow-up, the patient was relieved of the symptoms without any complications, and the wound healed well.

DISCUSSION

Paraurethral leiomyomas constitute a rare entity. They are usually found in females with a mean age of 40–44 years and are believed to be hormone-dependent. However, cases have been reported in females as young as 19 years till even in postmenopausal women [2,3]. Embryologically, they are believed to originate from the mesenchyme surrounding the urogenital sinus.

The spectrum of presentation may vary from asymptomatic cases to patients with voiding complaints, dyspareunia, or mass per vagina. The hormonal regulation is also corroborated by reports of variations in size during pregnancy followed by postpartum regression in size [4].

Due to the anatomic proximity, there may be a diagnostic dilemma between urethral, paraurethral, and anterior vaginal wall leiomyomas. However, the differentiation may help in guiding further management. Urethral leiomyomas may present with hematuria and may be fixed on examination as opposed to paraurethral leiomyomas which are generally mobile. Cystourethroscopy is a valuable adjunct for the diagnosis. The distinction between the two becomes important for the management as urethral leiomyomas are generally resected by the transurethral route, whereas the transvaginal approach is preferable for paraurethral leiomyomas. The other differential diagnosis to be considered include urethral diverticulum, vaginal cysts, urethral caruncles, Skene gland abscess or cyst, mucosal prolapse, and Gartner's duct cyst [5].

Transvaginal USG and MRI play a valuable role in the diagnosis. They help to identify the organ of origin and characteristics of the lesion and rule out any infiltration into the surrounding structures.

MRI can also exclude diverticula. Furthermore, USG can be used to guide biopsy in doubtful cases [6]. Paraurethral leiomyomas appear as well encapsulated hypoechoic homogenous masses on USG whereas on MRI, they may be hypointense or isointense on T1 and hyperintense on T2.

In case of doubt regarding the diagnosis, a pre-operative biopsy may be considered. Surgical excision is the management option of choice. The transvaginal route is the most preferred access route. However, the abdominoperineal approach has also been described for large lesions. In case of large tumors, there have been some suggestions for pre-operative embolization and treatment with gonadotropin-releasing hormone to reduce tumor size and intraoperative blood loss [7]. The main challenge during surgical excision is to prevent damage to the urethral sphincter and to maintain the adequacy of the periurethral support while simultaneously ensuring complete excision of the mass. The complications include injury to the bladder neck and injury to the urinary sphincter at the mid urethral level which may lead to incontinence, loss of periurethral support, and urethrovaginal fistula. The distal urethra contains the compressor urethra muscle, urethrovaginal sphincter muscle, and bulbocavernosus muscle, all of which should be spared with careful dissection [8]. Recently, a lateral transvestibular approach has been described. It offers the advantage of reducing the risk of urethrovaginal fistula by preserving the vaginal mucosa, along with improved visualization of periurethral structures [9]. In cases where hysterectomy is also planned concomitantly, the procedure can be done with robotic-assisted laparoscopic hysterectomy and mass excision with the anterior approach as it is less invasive and helps in better visualization [10]. Migliari *et al*. proposed adding a supportive fascial sling if the urethral meatus is involved. Rehabilitation should be started, and surgical management should be delayed for 6 months in cases of stress incontinence [11].

Histopathology is the gold standard for the confirmation of the diagnosis. It shows positive staining for desmin, smooth muscle antigen, as well as, estrogen and progesterone receptors [12].

Although the tumor is usually benign, recurrences have been described in four cases till date in the literature [13]. The coexisting uterine fibroids, as in this case, are also a rare occurrence with only a few cases reported, although the clinical significance of this is still unclear [14,15].

CONCLUSION

Paraurethral leiomyomas are infrequent tumors arising from the periurethral smooth muscle and are an important differential diagnosis for paraurethral masses in females of reproductive age group. In view of the limited experience with this entity, every case provides significant insight for its diagnosis and management.

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