



Case report

Long-term follow up of paraurethral leiomyoma: A case report and literature review

Jawaher Alsahabi^{a,b,c,d,*}, Asmaa Benawadh^e, Elham Bamanie^{a,b,c,d}^a Urogynecology and Reconstructive Pelvic Surgery, King Abdulaziz Medical City, Riyadh, Saudi Arabia^b King Abdullah International Medical Research Center, Ministry of National Guard -Health Affairs, Riyadh, Saudi Arabia^c King Saud bin-Abdulaziz University for Health Science, Ministry of National Guard - Health Affairs, Riyadh, Saudi Arabia^d King Abdulaziz Medical City, Ministry of National Guard - Health Affairs, Riyadh, Saudi Arabia^e Obstetrics and Gynecology King Faisal Specialist Hospital, Riyadh, Saudi Arabia

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ABSTRACT

Background: Paraurethral fibroid is a rare condition. To date, there is no unified protocol to guide long-term follow up in this condition. This study reviewed case reports and summaries published in the last 10 years and focused on the management plans and follow up of patients with paraurethral fibroids.

Case presentation: We report the case of a 44-year-old woman who presented with urinary symptoms. Clinical examination and magnetic resonance imaging revealed an approximately 3 × 3 cm mass which was considered as a paraurethral fibroid. Complete surgical excision was performed. The patient was discharged with no post-operative complications. Histopathology confirmed the diagnosis of a benign leiomyoma.

Conclusions: Paraurethral fibroid is a rare condition which can be diagnosed with a high level of certainty based on clinical examination and imaging. A multidisciplinary team which includes experienced radiologists and urogynecologists or urologists who have the expertise to perform vaginal surgery with urethral reconstruction is essential for definitive management. Long-term follow up for expected possible complications is advisable. Further research with a larger number of cases is needed to recommend an evidence-based protocol for management of paraurethral fibroid.

1. Introduction and importance

1.1. Background

Urethral and paraurethral fibroids are rare benign mesenchymal tumors that originate from smooth muscle surrounding the female urethra [1–3]. Although there is no identified age group that is prone to these fibroids, they are often reported in the reproductive age [4]. As this condition is extremely rare, it is difficult to evaluate its true incidence [5]. However, some studies estimated the incidence to be approximately 5% of all paraurethral masses, which are present in 1 out of 1000 women [6]. In existing literature, urethral and paraurethral leiomyomas are not clearly distinguished from each other. Ozel and Ballard clarified the differences by proposing three characteristics: 1) mobility of the mass (paraurethral tends to be mobile); 2) compared with a paraurethral fibroid, a urethral mass tends to protrude through the urethral meatus third is relation to the urethra, which can be

identified during dissection [7]. Braga et al. hypothesized that a paraurethral fibroid originates from residual blood vessels and smooth muscle in an embryologic state [8]. Few reports highlighted the link between reproductive hormones and tumor size; tumors may decrease in size in menopause and postpartum periods, and increase during pregnancy [9]. Immunohistological studies have revealed estrogen and progesterone receptors in the tissue. However, some authors argued that the tumors have been found in postmenopausal women, suggesting no correlation with hormonal factors [4]. Wyman et al. reported that some paraurethral tumors were non-malignant [10].

The aim of this report is to present our case of a 44-year-old woman with a paraurethral fibroid. In addition, we review relevant literature on paraurethral fibroids.

2. Case presentation

A 44-year-old woman, medically and surgically free. Presented with vaginal discharge associated with urinary frequency and mild stress

* Corresponding author at: Urogynecology and Reconstructive Pelvic Surgery, King Abdulaziz Medical City, Riyadh Sample University/Hospital, Saudi Arabia.
E-mail address: alsahabija@gmail.com (J. Alsahabi).

List of abbreviations

MRI	magnetic resonance imaging
US	ultrasound
CT	computed tomography
GNRH	gonadotropin-releasing hormone
BOO	bladder outlet obstruction
PVR	ost void residual urine test
IR	interventional radiology

incontinence. Obstetric score was G8P8 + 0 with all term pregnancies; births were via vaginal delivery without complication. Menstrual cycle was regular with normal volume and cycle length. The patient was not on contraception. On physical examination, a firm, nontender paraurethral mass measuring approximately 3 × 3 cm was noted to be pushing the urethra to the left. No cystocele was noted. Other medical history and remaining physical examination were unremarkable. As the concern was related to soft tissue pathology, Magnetic resonance imaging (MRI) showed a 2.7 × 2 cm homogenous right paraurethral mass (Fig. 1). The patient was managed as a case of complicated periurethral cyst/diverticulum, with the presence of fibroids considered. In addition, two small uterine fibroids were found incidentally, which were unlikely to be malignant.

A multi-disciplinary team composed of urogynecology, urology, and radiology services discussed the differential diagnosis. A pelvic examination was performed under anesthesia, which showed that the mass had a solid consistency and no communication with the urethral meatus. Complete excision was performed in the following surgical technique: complete vaginal excision of paraurethral mass. A Foley catheter was inserted into the urethra. Complete enucleation was performed. A simple interrupted suture was placed using absorbable sutures to close the dead space and the vaginal mucosa. Cystoscopy confirmed an intact urethra, bladder, and ureteric orifices. The catheter was removed. Estimated blood loss was 20 mL. The patient was observed post-operatively, and then discharged after regaining the ability to void spontaneously in the same day.

Histopathologic studies confirmed a benign leiomyoma (Fig. 2), with the widest diameter of 3 cm. The lesion was negative for malignancy. The postoperative period was unremarkable. The patient was followed up for two years. In the interim, the patient remained asymptomatic

without bladder symptoms or mass recurrence.

3. Discussion

Based on our review of existing literature, paraurethral fibroids have been reported to have a variable presentation (Table 1). This variety may be related to the size of the mass or relationship with the urethra. Approximately 50 % of the cases were asymptomatic [4]. Some patients presented with symptoms usually related to mass effect on the urogenital structures (e.g., recurrent urinary tract infections, voiding dysfunction or dysuria, abdominopelvic heaviness, vaginal bleeding, or hematuria) [1,5,13]. Urine retention is uncommon. However, Chodisetti et al. reported a case of urine retention due to a large paraurethral fibroid (8 cm). In 80 % of the cases, paraurethral leiomyomas appeared smooth and round, with solid consistency and sizes ranging from 1 to 8 cm [5,15].

Female paraurethral masses are rare. Differential diagnoses to consider include urethral prolapse, diverticulum, urethral caruncle, Skene's duct cyst, Gartner's duct cyst, Müllerian remnant cyst, epithelial inclusion cyst, ectopic ureterocele, congenital paraurethral cyst, vaginal neoplasm, fibrous polyps, urethral carcinoma, and mesenchymal tumors [11].

Despite the rarity of this condition, diagnosis can be established preoperatively through clinical examination and appropriate imaging modalities, including vaginal or transperineal ultrasonography (US), pelvic MRI, voiding cystourethrography, and computed tomography (CT) [11]. Ultrasonography and MRI can provide information on mass consistency and components, location, and possible features of malignancy. MRI can also exclude diverticula. Furthermore, US can be used to guide biopsy if there is suspicion of malignancy [1,12].

Surgical planning is fundamental, which requires detailed knowledge of the location of the mass and the anatomy of the female urethra to facilitate reconstruction. Preoperative distinction between urethral and paraurethral fibroids aids in surgical planning and patient counselling regarding postoperative management and expected outcomes [13]. Imaging modalities, such as US and MRI, may identify the exact location of a urethral leiomyoma, its morphologic and structural characteristics, the anatomic relationships between the mass and surrounding structures, the depth of tissue infiltration, vascularity, and signs of malignancy [12].

Leiomyosarcomas were not reported in similar conditions. High-signal intensity on T2-weighted MRI, ill-defined borders, and heterogeneous appearance on imaging are suspicious for malignancy.

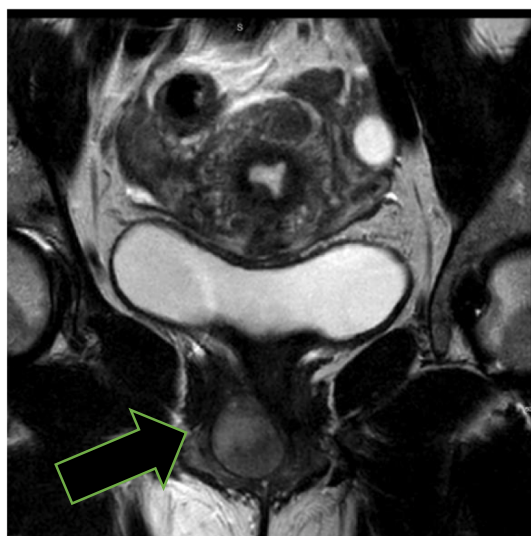
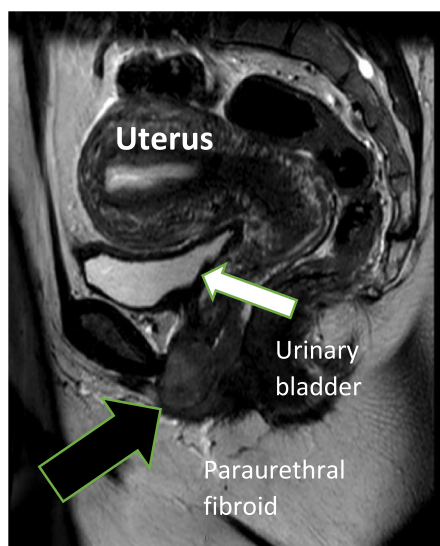


Fig. 1. Sagittal view of magnetic resonant Image (MRI) black arrows represent paraurethral fibroid.

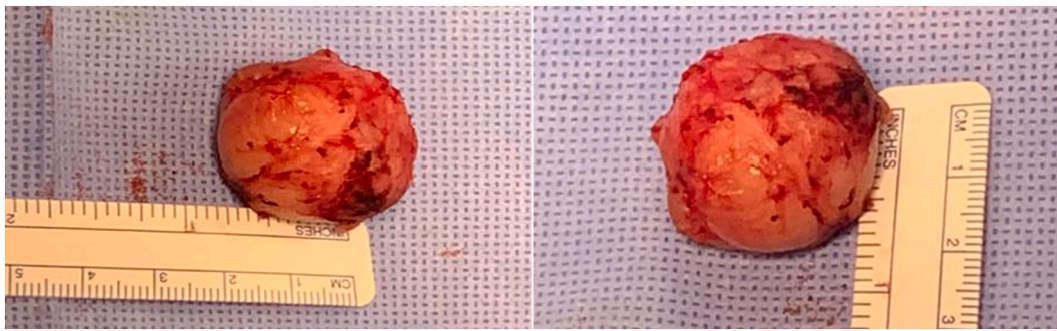


Fig. 2. Excised Paraurethral mass.

Clinically, rapid growth, an infiltrative nature, and the presence of metastases increase the risk for malignancy of a lesion [16]. Due to the potential effect of hormones on the lesion, medical management with gonadotropin-releasing hormone (GnRH) agonist has been proposed [1]. Twenty-five cases of paraurethral fibroid reported in the last 10 years were all treated surgically. A vaginal approach was chosen for complete excision, which offered symptom relief and allowed histopathologic studies to be conducted. Biopsy was required to exclude malignancy [2]. In the case reports, the most common intraoperative complications encountered were bladder injury (in two cases) and immediate postoperative vesicovaginal fistulae (22 cases) [4]. The latter might be related to the diverticulum component of the mass and the excision technique in cases where urethral diverticulum is involved [1]. Another potential postoperative complication is urinary incontinence [1]. Migliari et al. proposed adding a supportive fascial sling after mass excision if the urethral meatus is involved. Rehabilitation should be started, and surgical management should be delayed for 6 months in cases of stress incontinence. MRI is used to confirm complete enucleation or excision and recurrence of the leiomyoma, as well as for urodynamic investigation to confirm the stress component of urinary incontinence [1].

Reviewed reports showed that postoperative catheterization is inconsistently performed between 1 and 12 days. For our index case, neither urethral diverticulum nor injury were encountered, thereby warranting immediate removal of the catheter after surgery. Follow up was continued for two years as late complications have been reported several months post-surgery [1].

To date, there has been no reported malignant transformation [1]. Recurrence is extremely rare. There were two reported cases of recurrent urethral leiomyoma. One was large; the other was deeply embedded in the urethra, suggesting that primary resection was incomplete. Shen and Yang [13] and Jiménez et al. [14] each reported one case of recurrent paraurethral fibroid. One case required multiple resections at 6 and 9 years after the primary resection. The mass was initially 7.25 cm in size [13]. The other case had a repeat surgery 8 years after the primary resection. The mass was small but adherent to the urethra [14]. For recurrent cases, Previous studies recommend performing an MRI to confirm the absence of malignancy and to assess the possibility of delaying surgical management for at least 6 months [1,14,15]. Finally we would like to confirm that the work has been reported in line with the SCARE 2020 criteria [26].

4. Conclusion

Paraurethral fibroid is a rare condition which can be diagnosed with a high level of certainty based on clinical examination and imaging. A multidisciplinary team including experienced radiologists and urogynecologists or urologists who have the expertise to perform vaginal surgery with urethral reconstruction is essential for definitive management. Long-term follow up for expected possible complications is advisable. Further research with a larger number of cases is needed to

recommend an evidence-based protocol for management of paraurethral fibroids.

Ethics approval and consent to participate

Written informed consent was obtained from the patient for publication of this case report and accompanying images. Ethical approval was waived by the IRB committee of King Abdullah International Medical Research Center (KAIMRC).

Consent for publication

The patient has provided written consent for the publication of this article. Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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JA collecting the data, reviewing literature and writing the manuscript.

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EB designing the paper, critical appraisal of literature review and finalizing manuscript.

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Table 1
Summary of paraurethral /urethral fibroid reported in the last ten years.

Author, Year, Reference no.	Age	Presentation	Size (mm)	Investigations	Catheter	Follow up
Roberto Migliari, et al. 2015 [1]	32	All patients complained of vaginal swelling,	22	MRI	10	Clinical examination, MRI at 6 months and 2 years PVR, and uroflowmetry at 1, 6, and 12 months: only in case of urinary incontinence
	49	BOO, dysuria, and a low urinary flow.	32	US-guided biopsy		
	35		37	Uroflowmetry		
	43		57	Cystoscopy		
	37		42			
	47		55			
Ko Harada, et al. 2018 [2]	50	A protruding mass at the urethral opening for three months	36	MRI	1	No follow up reported.
Subbarao Chodisetti, et al. 2015 [5]	25	Acute urine retention Paravaginal mass 2 years	80	MRI, transperineal US Cystourethroscopy	5	One year follow up, no complications
C. Altay, et al. 2017 [3]	53	Progressive dysuria and vaginal bleeding 4/12	3	MRI ultrasound	N/a	7/12 asymptomatic
Dmitry Fridman, et al. 2018 [6]	52	Incidental	5	MRI, IR biopsy	7	
Giacomo Perugia, et al. 2012 [16]	25	Dysuria, dyspareunia, and obstructive voiding symptoms occasional urinary retention	2.5	Ultrasound, voiding cytogram, MRI	N/a	1/12 urinary flow evaluation performed 1 month after surgery revealed a normal peak urinary flow nomogram and the total absence of dyspareunia
Gamal Ghoniem, et al. 2016 [17]	20	Change in urine stream with intermittent spraying of urine	5	Ultrasound, MRI, cystourethroscopy and biopsy	8	Increased urinary frequency and occasional urgency, period is not available
Susy Shim, et al. 2015 [4]	59	Occasional incontinence, alternating with difficulties emptying the bladder	3	Ultrasound MRI	N/a	Immediate post op period vesicovaginal fistulae developed with reported improvement after repair
Shafqat Mukhtar, et al. 2011 [15]	23	Painless mass, severe dyspareunia, dysmenorrhea and vaginal discharge, difficulty in micturition with a poor and misdirected stream 2/12	6	Pelvic US micturating cystourethrogram	3 12	3/12 follow up: no complication apart from immediate post-operative urine retention
Kansal Jagan K, et al. 2016 [18]	49	Urinary frequency, urgency, nocturia and incomplete emptying with straining	3	MRI Biopsy	7	2/52 follow-up, asymptomatic
Soumya Cicilet, et al. 2016 [19]	28	Voiding difficulty with straining on micturition for 2 months	2.5	Uroflowmetry Transperineal us MRI	N/a	On follow-up, asymptomatic
S.V. Popov, et al. 2021 [9]	42	Frequent urination and urgency	3	Transperineal (US)	10	Asymptomatic; follow-up period is not available
Ritu verma, et all 2014 [25]	38	Enlarging perineal mass and dyspareunia for three years		Transperineal and transvaginal USG Cystourethroscopy MRI	25	The postoperative course was uneventful.
Amine Slaoui, et al. 2015 [21]	52	Dysuria and urinary tract infection and feeling of nodulation in her vagina	2,5		2	Asymptomatic; follow-up period is not available
Paolo Fedelini, et al. 2018 [22]	40	Hematuria, dysuria, recurrent urinary tract infections and dyspareunia	6	(USG) MRI urethroscopy	7	Followed-up for 6/12; no issues
Xia Wang, et al. 2022 [23]	44	Frequency, urethral meatus mass,	2.5	Ultrasound	N/a	Not available
	48	dysuria anterior vaginal wall mass,	4			
	36	dyspareunia anterior vaginal wall mass	2.5			
Marjon D. Viester, et al. 2020 [24]	28	Urinary frequency and urgency	3.7	Cystoscopy MRI	N/a	symptoms have resolved.
Junior de Lima, et al. 2014 [20]	52	Hematuria, dyspareunia, and feeling of nodulation in the vagina for 2 years	2	Flexible cystoscopy	10	Symptoms resolved

BOO, bladder outlet obstruction; PVR, post void residual urine test; MRI, magnetic resonance imaging; US, ultrasound; IR, interventional radiology; USG, NA, not available.

Declaration of competing interest

The authors have no competing interests to declare.

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