

A bladder leiomyoma masquerading as a vaginal mass causing dyspareunia. A case report and literature review

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ABSTRACT

Leiomyoma arising from the bladder wall are very rare yet benign tumors. We present a case of a 42-year-old female patient who presented to our unit with pelvic pain, dyspareunia and a feeling of “something coming down”. Following the utilization of a variety of diagnostic techniques (i.e., clinical examination and operative cystoscopy), a large fungating tumour measuring 5 cm was discovered. A vaginal approach was adopted to resect the mass. Histological and immunohistological examination of the tumor confirmed a bladder leiomyoma. Despite the resection being complicated by a cystotomy, the patient had an uneventful recovery and was completely asymptomatic at her subsequent follow up appointment. Given the rarity of this condition, we discuss the available literature on this unusual diagnosis.

KEYWORDS

Leiomyoma, bladder, benign, tumour.

Introduction

Leiomyoma of the bladder accounts for 0.43% of all bladder tumors^[1]. Bladder leiomyomas are the most common benign bladder tumors, yet they’re still very rare. Even though the majority of bladder tumors are epithelial in origin; however, leiomyoma originate from the mesenchyme^[2].

Bladder Leiomyoma can cause symptoms like dyspareunia, voiding dysfunction, and irritative symptoms like urgency and frequency^[3]. This usually depends on their location and size. They can also be asymptomatic and only discovered incidentally when doing a pelvic ultrasound or MRI.

There are about 250 cases of bladder reported in the literature and typically they are symptomatic with urinary voiding symptoms such as hematuria, irritative symptoms or urinary obstructive symptoms^[4]. We outline the case of a bladder leiomyoma in a woman who presented with dyspareunia. Owing to its’ rarity, diagnosis of a bladder leiomyoma was only confirmed following both histological and immunological examination.

Case report

A 42-year-old woman, was referred by her primary care physician to our unit with dyspareunia. She had two normal vaginal deliveries. Of note, she has no significant past medical or surgical history. Her symptoms started 3 months prior to her attending her primary care physician. She denied any urinary symptoms like frequency, urgency, nocturia, dysuria, hematuria, voiding difficulties or post void dribbling. She also

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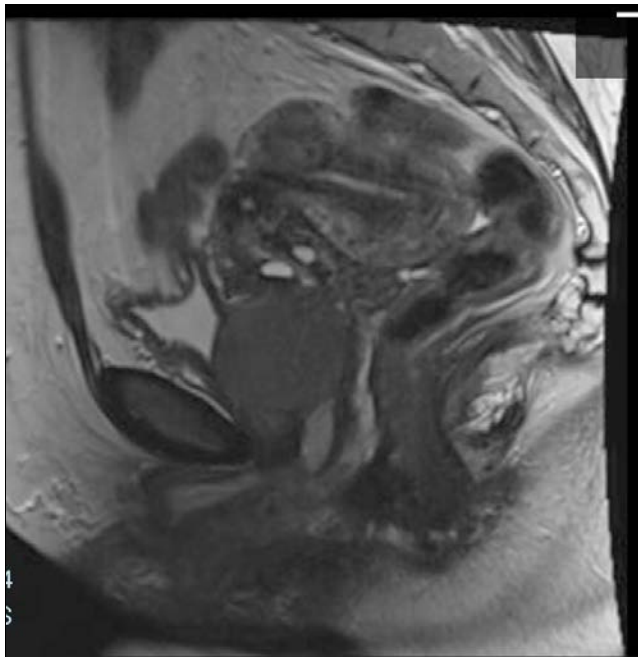
denied fever, chills, rigors or any symptoms of recurrent urinary tract infections. On examination, cervix appeared normal and a small amount of physiological vaginal discharge was noted. However, a mobile tender mass measuring about 4.4 cm was detected on the anterior vaginal wall. A pregnancy was ruled out and a urinalysis was normal. Her smears were up-to-date and have never showed any abnormalities. She also had a mirena coil in situ at the time of her review in clinic.

Following a multidisciplinary team discussion, a magnetic resonance imaging (MRI) was performed, confirming the presence of a large rounded mass measuring approximately 4.5 cm projecting from the anterior portion of cervix/upper vagina, demonstrating low/intermediate T1 and T2 signals with some mild enhancement post-gadolinium. The T2 MRI low signal showed the mass displacing the urethra anteriorly and vagina posteriorly at the level of trigone (Fig. 1).

Following discussion of the various treatment options with the patient, including transurethral resection, vaginal resection, and cystectomy the patient opted for vaginal resection. A vaginal approach allowed the proper identification of the entire mass. Due to the depth of bladder involvement, the identification plain was difficult.

This necessitated piecemeal removal of the specimen. At the time of dissection, a bladder injury occurred and was re-

Figure 1 T2 MRI low signal showing the bladder mass displacing the urethra anteriorly and vagina posteriorly at the level of the trigone.



paired in two layers. Bladder integrity was confirmed via cystoscopy and methylene blue instillation. The vaginal incision was closed in usual fashion using 2-0 serapide in continuous non-locking fashion.

The lady was discharged on the second post-operative day with an in-dwelling catheter. Prophylactic oral antibiotics were commenced. She was reviewed two weeks later for removal of the catheter and a trial of void. She also had a normal micturating cystourethrogram (MCUG).

She was reviewed at 6 weeks and 3 months postoperatively, and described complete resolution of her symptoms.

Microscopic results of histology using different levels of magnification with hematoxylin and eosin (H&E) staining showed a lesion made up of interweaving fascicles of spindle cells with elongated cigar-shaped nuclei (Fig. 2). Although vessels were easily identifiable with some extravasated red blood cells, no significant inflammatory component was revealed. Mitotic figures or necrosis were not easily identified. The histological features were in keeping with a leiomyoma, however, due to the unusual location of the lesion the differential diagnosis included fibromatosis, an inflammatory myofibroblastic tumour and gastrointestinal stromal tumors (GIST). Immunohistochemistry was carried out with tumour cells having strong and diffuse positivity for desmin and smooth muscle actin (SMA) and focal positivity for vimentin. The c-kit proto-oncogene product (CD117), anaplastic lymphoma kinase (ALK) and beta-catenin had negative staining while cluster of differentiation 34 (CD34) identified vessels within the tumour only. Both histological features and immunohistochemistry confirmed the diagnosis of a leiomyoma.

Within the specimen, overlying squamous epithelium was present, although the particular site of origin was still uncertain. The final diagnosis after a multi-disciplinary review of the case utilizing radiological features identifies this as a vaginal-vesical mass with characteristics of benign leiomyoma (Fig. 3).

Figure 2 Hematoxylin and eosin staining results of histopathological examination of bladder leiomyoma. (A) H&E stain; (B) desmin; (C) smooth muscle actin (SMA).

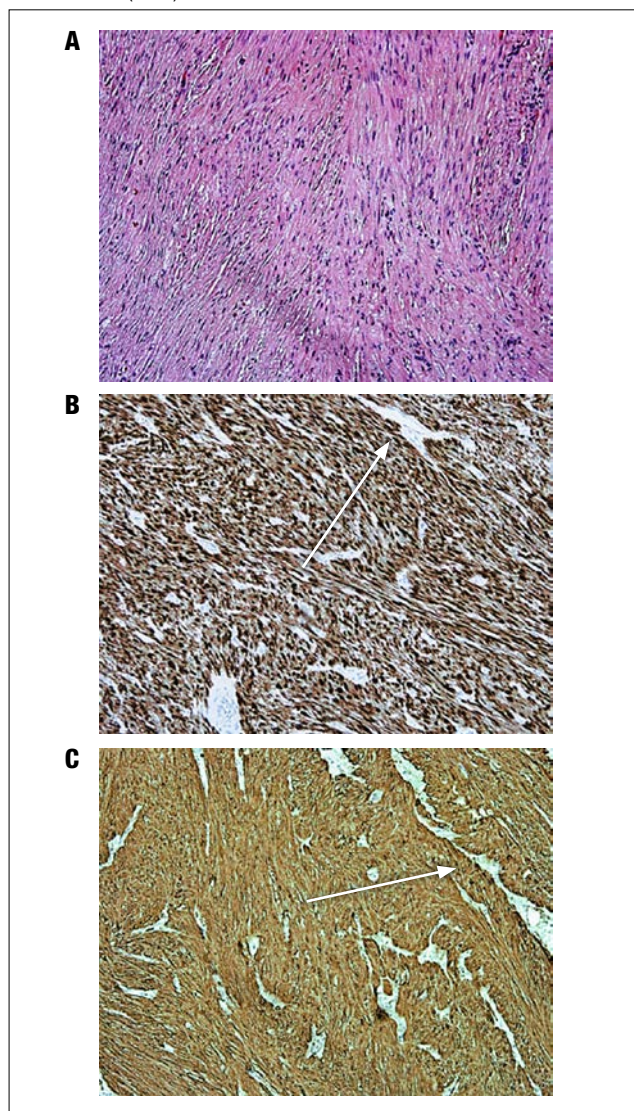


Figure 3 Gross appearance of the bladder leiomyoma mass postoperatively. Disintegrated fungating mass measuring 4.5 cm in length.



Discussion

We describe the unusual case of a bladder leiomyoma which presented with dyspareunia and a vaginal mass. Leiomyomata is mostly diagnosed among middle-aged females^[5] with a mean age of diagnosis estimated to be 45 years^[6]. The first case of bladder leiomyoma was reported in 1931 by Virchow^[7]. The patient had a large fungating bladder leiomyoma which had undergone mitosis thereby making this presentation an increasingly rare finding. Leiomyoma can develop anywhere within the genitourinary tract. Bladder leiomyoma mainly develop on the submucosa but can arise from any layer of the bladder, translating to three types of bladder leiomyoma, intravesical (63-86%), extravesical (11-30%), intramural (3-7%)^[8].

Women are disproportionately diagnosed with bladder leiomyoma compared to men with Dodia *et al.* accentuating that the incidence among women is three-fold higher than in men and mainly occurs at the age of 40s and 50s^[6]. There are a number of explanations regarding the aetiology of these, such as monoclonal cells arising from the endometrium^[9], or there being a hormonal influence^[4], explaining higher incidence following menopause^[10]. This explains the higher risk for developing bladder leiomyoma among women aged at 40s and 50s. Alternative explanations include Blum's irritative theory (relating to chronic inflammatory stimuli), and Piegel's disontogenic theory (embryological associations)^[11].

Leiomyoma of the bladder may be classified as symptomatic or asymptomatic. Symptomatic bladder leiomyoma is mainly identified by lower urinary tract symptoms such as pyuria, hematuria, irritative symptoms such as nocturia, urinary urgency, bladder discomfort, painful voiding, or stranguria, and urinary obstruction symptoms. Moreover, the literature identifies dyspareunia as a rare symptom in bladder leiomyoma^[7].

The severity of leiomyoma symptoms depends on the size as well as the location of the tumours^[11]. Larger leiomyomas present with more severe symptoms such as urinary retention and pain. However, Agrawal *et al.* provided a different line of argument in their findings which indicated that smaller bladder leiomyoma masses with a diameter less than 1.4 cm could cause symptoms such as urine retention and pain^[12].

Diagnosis of bladder leiomyoma can be achieved through ultrasound, ultrasonography, computerized tomography (CT) scan, or MRI whose results reveal homogenous smooth lesions accompanied by peripheral hyperechogenicity. Both ultrasound and ultrasonography can reveal presence of a solid mass on bladder submucosa as well as confirm the origin of the tumour. Both ultrasound and ultrasonography are thus essential in the differential diagnosis of bladder leiomyoma as they can rule out diagnoses of cervical fibroid^[13].

Both CT scan and MRI can help differentiate tumours based on their contents as well as their relationship to the neighboring structures. These techniques can identify the mesenchymal content of tumours with higher superior precision but are reportedly ineffective in differentiating mesenchymal tumours from the frequently identified transitional cell tumours^[5]. This makes cystoscopy and biopsy of lesion essential before leiomyoma is explored, especially in ruling out the presence of endovesical tumour^[12].

Due to the limitations of these imaging methods, histopathological study can help, confirm the diagnosis of bladder leiomyoma diagnosis, as Itam *et al.* explain^[14].

However, histopathological examination, in this case, could not confirm the diagnosis of bladder leiomyoma and immunohistochemistry coupled with H&E staining results can successfully diagnose bladder leiomyoma^[15].

There are various types of treatment and intervention for bladder leiomyoma including surgical methods such as open surgical excision and transurethral resection. Both of these methods can be reliably used in the treatment of small tumours and have been reported to have excellent prognosis^[15]. Other minimally invasive surgeries such as open cystostomy are recommended for symptomatic patients and can lead to excellent results without fear of recurrence^[16]. However, larger tumours may be treated effectively through open surgery with partial cystectomy or segmental resection^[11]. Other treatment options such as robotic laparoscopic resection of bladder leiomyoma have been successfully applied in the treatment of large bladder leiomyoma^[14] whereas vaginal resection is recommended in certain special cases to remove bladder leiomyoma^[11].

Conclusions

We present the case of an unusual vaginal mass causing dyspareunia. After excision, the final diagnosis was that of a bladder leiomyoma. Various imaging techniques can aid in the diagnosis of vaginal masses and bladder leiomyoma in particular, however the final diagnosis is always a histological one owing to the rarity of these masses. A vaginal approach to the resection has also shown to be a safe and effective treatment intervention for bladder leiomyoma without compromising bladder function.

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