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Paraurethral Leiomyoma during Pregnancy: A Case Report and Review of Literature

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Abstract

Leiomyoma is a benign tumor of smooth muscle cells that may arise from the genitourinary or gastrointestinal systems. It is not common to arise from the urethra or Para urethral areas with few reported cases. It is important to differentiate urethral leiomyoma that presents mainly with obstructive urinary symptoms and require surgical endoscopic resection from the para-urethral one that presents mainly as asymptomatic freely mobile lump and not communicating with the urethra with a clear line of cleavage in between. Review of literature showed that leiomyoma is hormone sensitive and progressively increase in size during pregnancy and may cause obstructed labor or obstructive urinary symptoms according to its site. We present a case of para-urethral leiomyoma during pregnancy presented with painless lump that progressively increased in size causing obstructive urinary symptoms. Examination under anesthesia was done and diagnostic cystoscopy-urethroscopy prior to surgical excision to ensure site of origin with no urethral communication. Histopathological evaluation confirmed the diagnosis of leiomyoma. No postoperative complications were noted with improvement of the urinary symptoms.

Keywords: Leiomyoma; Periurethral mass; Benign urethral tumors; Leiomyoma during pregnancy

Introduction

Leiomyoma is a benign lesion of the smooth muscle cell arising most frequently in the genitourinary and gastrointestinal tracts. The uterus is considered the most commonly involved in the genitourinary tract, but it could arise from other sites like renal pelvis, ureters and urinary bladder wall. Both urethral and paraurethral leiomyoma and considered to be extremely rare [1,2].

A true paraurethral leiomyoma unlike urethral leiomyoma has no connection to the urethral, vaginal or the urinary bladder. Like uterine leiomyoma they are hormonal dependent with potential malignant transformation [3]. Local recurrence may occur after complete excision, but no metastasis has been reported [4].

We present a case of paraurethral leiomyoma in a pregnant patient after confirming the diagnosis by a diagnostic cystoscopy and excision biopsy.

Case Presentation

A 25 years old female patient, presented to us with painless paraurethral swelling during her first trimester of pregnancy, this swelling showed a slowly progressive course during the second trimester, the swelling increased in size associated with obstructed urinary symptoms in the form on difficulty and straining during micturition, hesitancy, weak interrupted stream of urine and sense of incomplete evacuation, no other storage or voiding symptoms were mentioned by the patient. On examination a bean shaped painless soft swelling related to the upper border of the urethral meatus with no discharge from the swelling Figure 1.

Abdominal and pelvic ultrasound showed post voiding residual urine of 300 cc, with obstructed uroflow, we proceeded for examination under anesthesia that showed a freely mobile para-urethral soft swelling related to the upper border of the meatus, diagnostic urethrocystoscopy was done prior to excision that showed a mass compressing the urethral lumen with no communication, excision of the mass was done with ligation of the pedicle Figures 2 and 3.



Figure 1: The paraurethral swelling during initial examination under anasthesia.

Gross examination revealed a lobulated whitish soft mass of 3×2 cm (Figure 4). Immunohistochemistry confirmed the diagnosis of leiomyoma with immunoreactivity for estrogen receptors. No postoperative complication was noted during the patient follow up till 6 months postoperative.

Discussion

Leiomyoma is a mesenchymal benign tumor arising from smooth muscle fibers, distinction between urethral, Para urethral and vaginal leiomyoma is clinically challenging this is due to the anatomical adherence and they share a common presentation. Urethral leiomyomas

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Figure 2: After excision of the liomyoma in relation to the uretheral meatus.



Figure 3: Excision of the mass with good hemostasis after ligation of the pedicle.

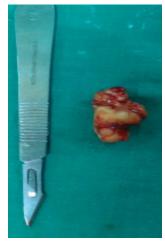


Figure 4: Gross examination revealed a lobulated yellowish soft mass of 3 × 2 cm.

most commonly occur at the posterior wall while vaginal one usually arises from the anterior wall and Para urethral leiomyomas arise from the scarce tissues in the Para urethral space [5,6].

Clinically leiomyoma may be present as protruding asymptomatic mass or may be presented with dysuria, hematuria, dyspareunia, obstructive urinary symptoms or even urinary retention or even may be encountered accidently during surgical dissection during incontinence surgical procedure. The average age of presentation usually between 40 and 44 years [6-10].

Peri-urethral masses include a variety of benign and malignant lesions including, urethral caruncle, skene's gland abscess or cyst, mucosal prolapse, ectopic ureterocele, urethral diverticulum, vaginal wall cyst, Gartner's duct cyst, hamartoma, leiomyoma, carcinoma or sarcoma, where urethral diverticulum, leiomyomas and skene's gland cyst represent the 3 most common causes of these masses [2-12].

Although there is no clear consensus on the most accurate diagnostic investigation, reports suggest transvaginal ultrasound shows leiomyomas as solid tumors with a homogeneous internal echo structure, it can rule out infiltration and differentiate them from diverticula, and can determine their relationship with respect to the urethra [13,14]. Voiding cysto-urethrogram (VCUG) helpful to evaluate the urethral and any lesion that may arise from it or diverticulum and possible communication of a para-urethral lesion [7], magnetic resonance imaging (MRI) shows a signal intensity in T1-weighted sequences analogous to that of pelvis muscle, while in T2 sequences an intensity similar to fat, referring to a solid mass [1,13] or visualized urodynamic study (VUDS) especially in cases presenting with lower urinary tracts symptoms, biopsy were obtained prior to excision was reported for the suspicious of malignancy [7]. Diagnostic cystourethroscopy remains a corner stone in management of such cases especially in differentiating urethral from paraurethral leiomyomas.

Although there is no clear pathogenesis of leiomyomas, reports have documented hormonal dependence to estrogen and variation in size during pregnancy like our case and postpartum regression in size that was documented by estrogenic receptors staining by immunohistochemistry [15-18].

Unlike urethral leiomyoma where surgical excision is the treatment of choice, surgical intervention is usually not needed for asymptomatic para-urethral leiomyoma for the fact that no malignant transformation has been reported [17,19,20], in addition to the potentiality for spontaneous regression as being a hormonal dependent. However, we believe that surgical resection and histological diagnosis is the best of choice for accurate diagnosis of leiomyoma and to distinguish it from other malignant lesions also some of them may not regress or even cause later symptoms. Few cases of paraurethral leiomyoma during pregnancy has been reported where symptoms varies according to tumor size from asymptomatic vaginal lump up to difficult obstructed labor and a case has been reported for ruptured uterus from a huge vaginal mass where surgical excision is the treatment of choice during the second trimester [21-23].

Malignant leiomyosarcoma as reported by Tavassoli and Norris should be suspected with a lesion more than 5 cm in size with more than 5 mitotic figures per 10 high power field, infiltrative margins [24] although no report cases of sarcomatous changes in the literature. A vaginal approach is used to address vaginal and paraurethral leiomyoma, while transurethral resection is preferred for urethral one [19-25].

Although urethral leiomyoma is usually associated with urological

symptoms, it may be asymptomatic and to differentiate from paraurethral leiomyoma, diagnostic cystourethroscopy prior to surgical excision help is mandatory in such cases to clarify if there is a communication with the urinary system or not. What also aid in differentiating urethral from paraurethral leiomyoma is that paraurethral leiomyoma is usually mobile not fixed and of mesenchymal origin contained in a fibrous capsule not adherent or communicating with the surroundings and once it is removed there will be no mucosal disruption of the urethra [25-27].

Conclusion

Para-urethral leiomyoma is a benign lesion that may be present in a varies ways ranging from asymptomatic lump up to obstructive urinary symptoms or recurrent infection, during pregnancy it may be cause an obstructed labor, it shows hormonal sensitivity and it may increase in size during pregnancy and regress afterward. There is no clear consensus on the definite diagnostic modality. However, diagnostic urethro-cystoscopy prior to surgical excision is mandatory prior to excision to differentiate it from a urethral leiomyoma and ensure no communication with the urethra. We believe surgical excision is best of choice regarding management of such lesions and for accurate histopathological diagnosis.

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