Case Report

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Paraurethral Leiomyoma: An Extremely Rare Extrauterine Leiomyoma

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ABSTRACT

Leiomyomas occur most frequently in the genitourinary and gastrointestinal system. Amongst all the leiomyoma cases, the most common site of occurrence is the genital tract (95%), while the remaining tend to occur at various other sites of the body. However, paraurethral leiomyomas are extremely rare benign mesenchymal tumours and only few cases have been reported in the literature. Herein, we describe a case of paraurethral leiomyoma in a 29-year- old female. MRI revealed a well-defined signal soft tissue intensity lesion measuring approximately 2.67 SI x 2.3 AP x 1.9 TR cms, in the space between urethra and vagina, closely abutting both the structures. Mass was excised under spinal anaesthesia. Histopathological examination along with immunohistochemical positivity for smooth muscle antigen confirmed the diagnosis of leiomyoma.

Keywords: *Leiomyoma, Paraurethral Leiomyoma, Histopathological Examination*

Introduction

Paraurethral leiomyoma is a rare, benign hormone dependent neoplasm of mesenchymal origin affecting women of reproductive age group (fourth or fifth decade of life). Fifty percent of the patients are asymptomatic.[1] Urethral leiomyoma was first described by Buttner.[2] Paraurethral leiomyoma constitutes approximately 5% of all urethral tumours.[3]

It commonly arises from posterior upper urethra. Size ranges from few millimetres to few centimeters.[4] Only few cases of paraurethral leiomyomas have been documented in the literature out of all the reported urethral tumors.[5]

Herein, we describe a case in a 29-year-old female patient of reproductive age who presented with a mass at paraurethral region which turned out to be leiomyoma on histology.

Case Report

A 29 –year- old female presented with complaint of asymptomatic swelling in perineum for 3 years which was insidious in onset and progressive in nature. There was no history of frequency, urgency, voiding lower urinary tract symptoms or poor urine stream. Also, there was no history of hematuria, lithouria, fever and weight loss. Complete blood count was within normal range. KFT, LFT and routine urine microscopic examination didn't show any abnormality. The patient didn't have any significant medical disease or surgical history. The lesion was clinically suspected to be a urethral diverticulum. Further, radiological investigations were performed. Ultrasonography of whole abdomen revealed a well defined soft-tissue lesion at the level of lower 1/3 rd of urethra and vagina, suggestive of benign neoplastic lesion arising from posterior urethral wall and well-maintained fat plane of lesion with vagina. MRI pelvis showed a well-defined signal soft tissue intensity lesion in lower 1/3 rd of urethra and vagina measuring approximately 2.67 SI x 2.3 AP x 1.9 TR cms (Figure 1). It appeared hypertense on T2 and isointense on T1 weighted images. The lesion was lying in the space between urethra and vagina, closely abutting both the structures. The lesion didn't appear to be infiltrating urethra. Color doppler imaging revealed increased flow of blood. No evidence of cystic or calcific focus was seen within the lesion. No obvious surrounding fat stranding was seen.

Local examination showed a globular 2x3 cm swelling in posterior aspect of urethra at 6'0 clock position. Swelling was firm in consistency with well defined margins. Paraurethral mass excision was done under spinal anaesthesia. The exicision biopsy showed a grey-white tissue mass measuring 3 x 2 x 1.5 cm. Post operative period was uneventful without any complications.

On gross examination, the mass was globular, firm and congested with grey-white homogeneous cut surface. Microscopic examination revealed a tissue focally lined by parakeratotic squamous epithelial lining. Underlying subepithelium showed a tumour with cells arranged in sheets and interlacing fascicles (Figure 2). These tumour cells were oval to spindle with bland nuclear chromatin and moderate to abundant eosinophilic cytoplasm. There

was no mitosis, pleomorphism or necrosis (Figure 3). Immunohistochemistry (IHC) for smooth muscle antigen (SMA) showed diffuse positivity (Figure 4). Histological and IHC features were consistent with paraurethral leiomyoma.

Discussion

Leiomyomas are classified into following three categories—skin leiomyomas, angiomyomas (vascular leiomyomas) and leiomyomas of deep soft tissue. Urethral leiomyomas are categorised as deep-tissue leiomyomas. They are much larger as compared to their superficial counterparts and exhibit a greater spectrum of histological changes; therefore, it becomes very important to clearly distinguish them from leiomyosarcomas, which are also more common in deep soft tissue. In addition to this, leiomyoma of urethra is a misleading term, since they do not seem to arise from intraurethral muscular layer. Normally they originate from smooth-muscular component around urethra, thus they should be more correctly identified as paraurethral leiomyoma.[6]

Common complaints include a mass, urinary tract infection, dyspareunia, hematuria, urinary retention, dysuria and obstructive voiding symptoms.

On the contrary, patients can be completely asymptomatic as in our case.[7]

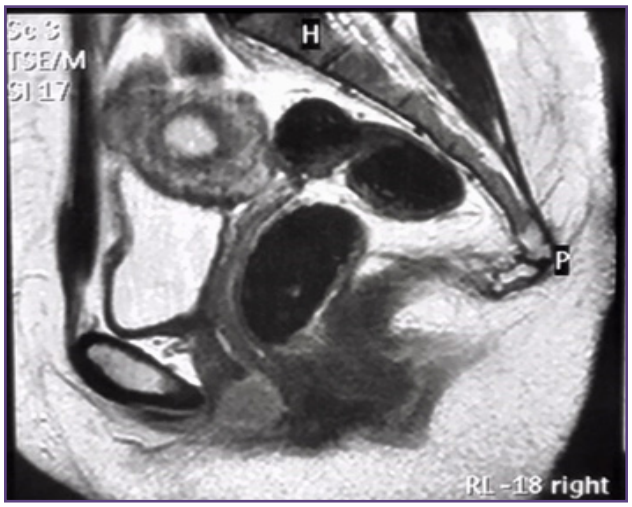

Fig. 1: Radiological image of the lesion.

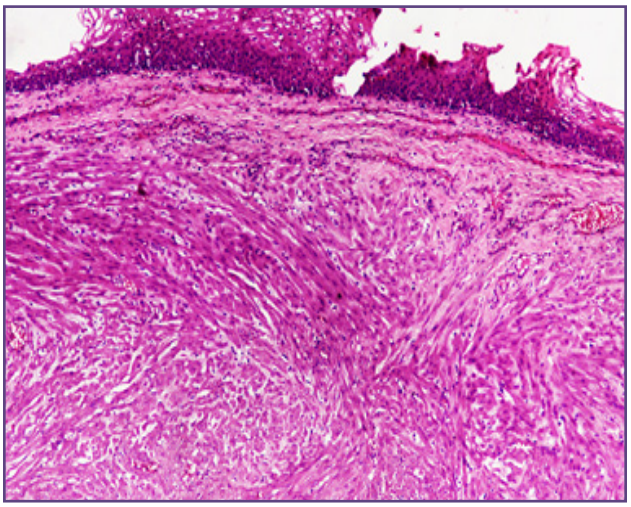

Fig. 2: (H&E 10X)- Shows a tissue focally lined by parakeratotic squamous epithelial lining. Underlying subepithelium shows tumour with cells arranged in sheets and interlacing fascicles.

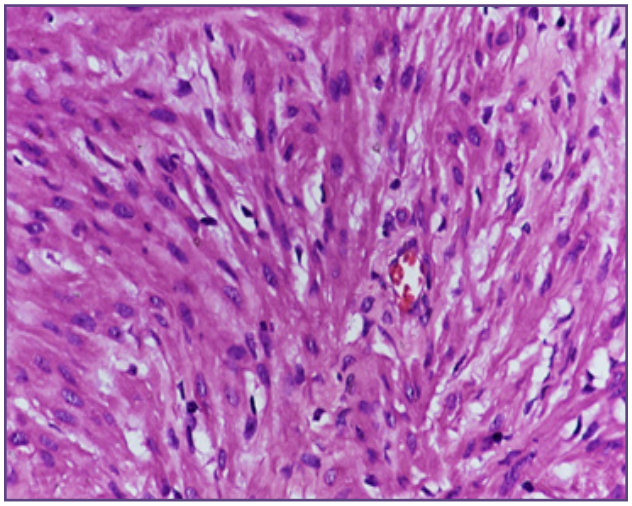

Fig. 3: (H&E 40X)- Tumour cells are oval to spindle with bland nuclear chromatin and moderate to abundant eosinophilic cytoplasm.

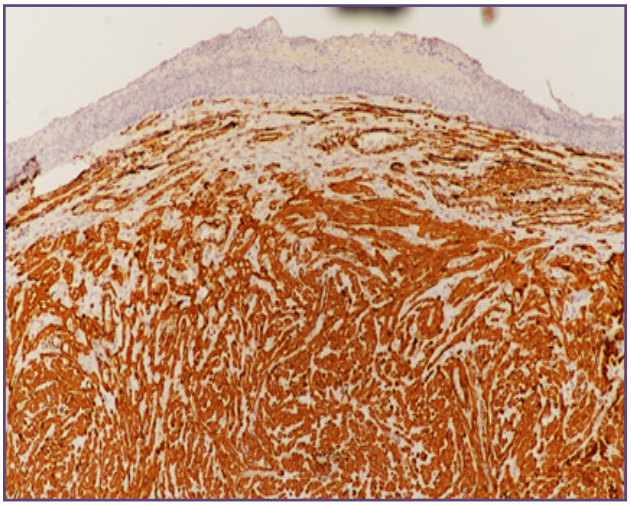

Fig. 4: (H&E 40X)- IHC for smooth muscle antigen (SMA) showed diffuse positivity.

Etiology of leiomyoma is unknown. The relatively increased incidence in reproductive age women in comparison to younger and older women has led to speculation that circulating female hormones lead to enhanced leiomyoma growth.[8]

Due to close anatomical positioning, distinction between vaginal, urethral and paraurethral leiomyoma is often very difficult. In addition, they have similar clinical presentation. Paraurethral leiomyomas have a tendency to occur between posterior urethral wall and anterior vaginal wall in the paraurethral space or vesicovaginal septum.[1]

On literature review, we could find 12 previously reported cases of paraurethral leiomyoma published in English language. The patients showed wide age distribution ranging from 16 to 64 years, with a mean age of presentation at 37.55 year.[9] The most common presentation was mass per urethra or vagina and obstructive lower urinary tract symptoms. All the cases were treated by surgical excision. Our case presented with mass per vagina, however she was otherwise asymptomatic.

Both USG and MRI provide important information pertaining to the diagnosis and extent of paraurethral leiomyoma. The relationship of paraurethral leiomyoma with respect to urethra can be determined by transvaginal USG. Paraurethral leiyomyoma appears as a well circumscribed, hypointense or isointense to muscle on T1WI and relatively low signal intensity on T2WI suggesting mesenchymal origin, on MRI. Irregular enhancement on post-contrast-enhanced images and heterogeneously enhancing lesions on T2-weighted MRI can point towards rhabdomyosarcoma and leiomyosarcoma.[9]

The various differential diagnosis includes congenital, cystic, inflammatory and neoplastic, vaginal and urethral conditions like urethral caruncle, prolapsed ectopic ureterocele, urethral diverticulum, Gartner's duct cyst, urethral carcinoma, papilloma, fibrous polyp. Rarely congenital paraurethral cysts, lymphoma, plasmocytoma, leiomyosarcoma, rhabdomyosarcoma and malakoplakia can also present as paraurethral mass lesions.[9] However, urethral leiomyoma is typically smooth, round and firm on gross examination with a size usually ranging from 1-8 cm in size and the final diagnosis is rendered on histological examination. [9, 10]

Tumour excision through vaginal approach is the treatment of choice in many previous reports.[5] However, due to close proximity to urethra and bladder a meticulous approach is utmost important to prevent injury during resection of the mass.

Conclusion

It is very important for the clinicians as well as pathologists to be familiar with differential diagnosis of lesions in the paraurethral region in order to achieve adequate management and favourable patient outcome. In order to demonstrate benign nature and site of origin of lesion, histopathological examination is considered imperative. The treatment of choice is surgical excision because it allows complete pathological examination so that any foci of malignant change is ruled out.

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Conflict of Interests

None declared

Ethics Statement Including Patient Consent

Approval from ethics committee is not required for a case report as per policy of our institute . A written informed consent was taken from the patient for use of her radiological and histopathological images for research or educational purpose.The study was conducted according to the principles of the Declaration of Helsinki.

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