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Extra-uterine Leiomyoma – Case of a Urethral Tumour in a 29-Year-Old Female

Urethral Leiomyoma in a 29-Year-Old Female

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Abstract : **INTRODUCTION**: Urethral leiomyoma is a rare benign tumour of smooth muscles origin occurring usually in women of reproductive age. This case is presented because it is the first seen in our practice of over two decades.
CASE PRESENTATION: We present the case of a 29-year-old para 0^{+1} female with complaints of vulva swelling and occasional urine dribbling. Clinical examination of the swelling of 5 years duration revealed a firm, 5 by 5cm lump that is attached to the urethra. The lump was excised and confirmed histologically to be urethral leiomyoma.
CONCLUSION: Leiomyoma in the urethra is a very rare but treatable condition. The gold standard diagnosis is routine histologic testing while ancillary investigations like USS, CT scan and MRI may be required for characterization of the tumour. Prompt intervention by surgical excision is sufficient in prevention of undue complications.

IndexTerms **– Leiomyoma, urethra, extra-uterine, tumour.**

INTRODUCTION

Leiomyomas have been classified under three categories: (i) cutaneous leiomyoma (leiomyoma cutis), (ii) angiomyomas (vascular leiomyomas), and (iii) leiomyomas of deep soft tissue.[1] Whatever category they belong, they are a benign smooth muscle tumour with very rare chance of malignant transformation. In our practice, they have been seen almost entirely in the uterus.
Urethral leiomyoma is a very rare tumour, arising from the smooth muscle layer of the urethra. They are more common in female than male.[2] Most reported cases were in the proximal segment of the urethra.[3] They are reported to be most common in women of reproductive age; with mean age of appearance put at around 41 years.[4]
Common presenting features may include recurrent urinary tract infection, presence of urethral mass, dyspareunia, dysuria, urinary retention, dribbling and urethral irritation.[2,5] If untreated, they can become a cause of bladder outlet obstruction with possible complications of hydroureter, hydronephrosis and renal failure ultimately. This is why they are important, beyond the discomfort associated with them.
A case of posterior urethral leiomyoma in a 29-year-old negroid female is being reported. The clinical diagnosis was that of a posterior urethral mass, query cause. The diagnosis was confirmed on histology following excisional biopsy which brought complete resolution of symptoms. This is the first such case seen in our practice of over two decades, hence the interest to report it and create awareness.

CASE PRESENTATION

Miss A.E was a 29-year-old para 0^{+1} unemployed lady with tertiary education. She presented to the gynaecology clinic of the Irrua Specialist Teaching Hospital, following verbal referral from a peripheral center, with complaints of vulva swelling of five years duration.

The swelling was said to have progressively increased in size and associated with occasional urine dribbling and staining. There was no vulva pain, fever, increased urinary frequency, urgency, nocturia, dysuria, stress urinary incontinence or swelling in other parts of the body. Even though she did not have dyspareunia, she shied away from sex.

At onset she thought it was part of normal growth of the vulva until she started having dribbling and staining. Following advice from friends she took several medications (orthodox and herbal), but without relief. This made her visit a peripheral center from where she was referred to us.

Vulva examination revealed a 5 by 5 cm, non-tender lump on the left introitus occluding the urethral opening. The solitary lump was firm and not attached to the vulva skin or mucosa. Its only attachment was in the urethra but there was no difficulty catheterizing her. She was subsequently subjected to routine pre-op assessments and scheduled for excision biopsy under loco-regional anaesthesia. The lesion was enucleated via a transverse 4cm incision over it, sparing the urethral mucosa. Surgical wound repair was done in layers, using absorbable sutures. The procedure was essentially uneventful with minimal blood loss. She was discharged home the same day on antibiotics and analgesics. When seen the following week at the clinic, the wound was healed satisfactorily, with no complaints.

Histology report described an encapsulated tumour composed of proliferating sheets of mature smooth muscle cells arranged in whorls and fascicles within scant intervening stroma (Figure 1). A diagnosis of urethral leiomyoma was made. Patient was reassured and placed on sexual abstinence for four weeks. She has since fully healed and resumed all her normal activities.

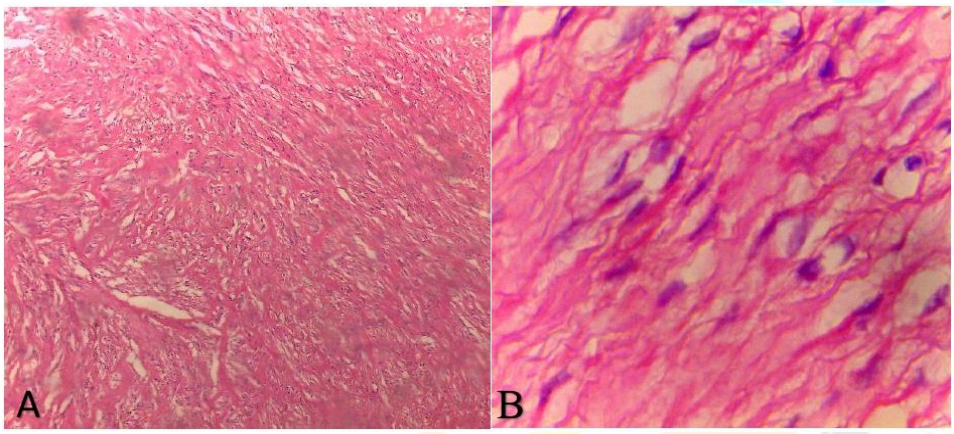

Figure 1: H & E Photomicrograph showing interlacing facscles of mature smooth muscle cells with pausity of mitotic figures.

DISCUSSION

Genitourinary tract leiomyoma makes up about 95% of all cases of leiomyoma reported in medical literature.[5] Despite being most common in the uterus, they have also occurred in uncommon sites and displayed unusual growth patterns in places like the vulva, ovaries, urethra, and bladder in the genitourinary tract.[6]

Urethral leiomyoma, which is categorized as leiomyoma of the deep soft tissues, is a very rare soft tissue tumour originating from the smooth muscle of the urethra.[7,8,9] It was first described in 1894 by Buttner as the most common mesenchymal neoplasm of the urethra.[5] According to Barut and Mohamud, urethral masses are uncommon, the benign of which include urethral caruncle, Skene's duct cyst, Gartner's duct cyst, papilloma, hemangioma, and leiomyoma.[6]

The urethra is a muscular tube that extends from the bladder neck and is composed of an inner layer of smooth muscle and an outer layer of striated muscle. The smooth muscle layer can be separated into an inner layer of longitudinally orientated smooth muscle and an outer, relatively thinner, layer of circular muscle.[10] The tumour arises from the circular fibres of the smooth muscle layer. No cause has been ascribed to their origin, but some authors link their pathogenesis to high levels of estrogen. They are said to achieve rapid growth and get bigger during pregnancy and to shrink with menopause.[11,12] This is not surprising because its counterpart in the uterus is well known to be estrogen responsive. It is likely though that beyond estrogen, other determinants may play roles in the pathogenesis. These other determinants are at present not defined. The role played by pregnancy or other factors in the present case could not be determined. The lady was a para 0^{+1}.

Like most other reported cases, the index case was in the posterior urethra. Though some of them have been reported to be symptom free, this one presented with few symptoms of swelling and dribbling. Some cases may unusually present with stress incontinence due to wide urethral dilation, requiring plastic urethral repair.[13] This patient was not incontinent for urine.

Clinically, the differential diagnoses often considered in such lesion as this include caruncle, diverticulum, urethral prolapse, papilloma, ectopic ureterocele, fibrous polyp, and even posterior urethral valve in the male. This is where histology becomes invaluable in clinching the diagnosis. Once diagnosis is confirmed, definitive treatment is surgical. The current case had an excision biopsy and needed no further treatment after diagnosis was confirmed.

The size of most urethral leiomyomas has been reported to range between 1 and 8 cm in diameter; the index case was 5cm, well within range. Leiomyosarcoma, a possible complication, was ruled out because of absence of atypia and significant mitotic activity. The tumour cells were spindled and arranged in whorls and fascicles within a capsule.

Urethral leiomyomas can be complicated with erosions, ulcerations or even malignant transformation, as is the case with other benign tumours. However, no malignant transformation has been reported to date concerning urethral leiomyoma. What has been seen is recurrence after surgical resection.[11,12] Female urethral leiomyoma could have social implications. In the index case there was avoidance of sex, possibly from discomfort or shame. Also, at onset of symptoms she thought it was part of normal growth of the vulva until she started having other symptoms. This happened because of poor education on basic female anatomy and contributed to delayed medical attention.

We conclude that this is a treatable condition. Prompt diagnosis and proper treatment are necessary to prevent complications.[14] We advocate renewed effort to meaningful education of the girl child which equips them with basic knowledge about their body structure and functions.

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