

## Female Urethral Cavernous Hemangioma, A Rare Entity: Two Case Reports and Review of The Literature

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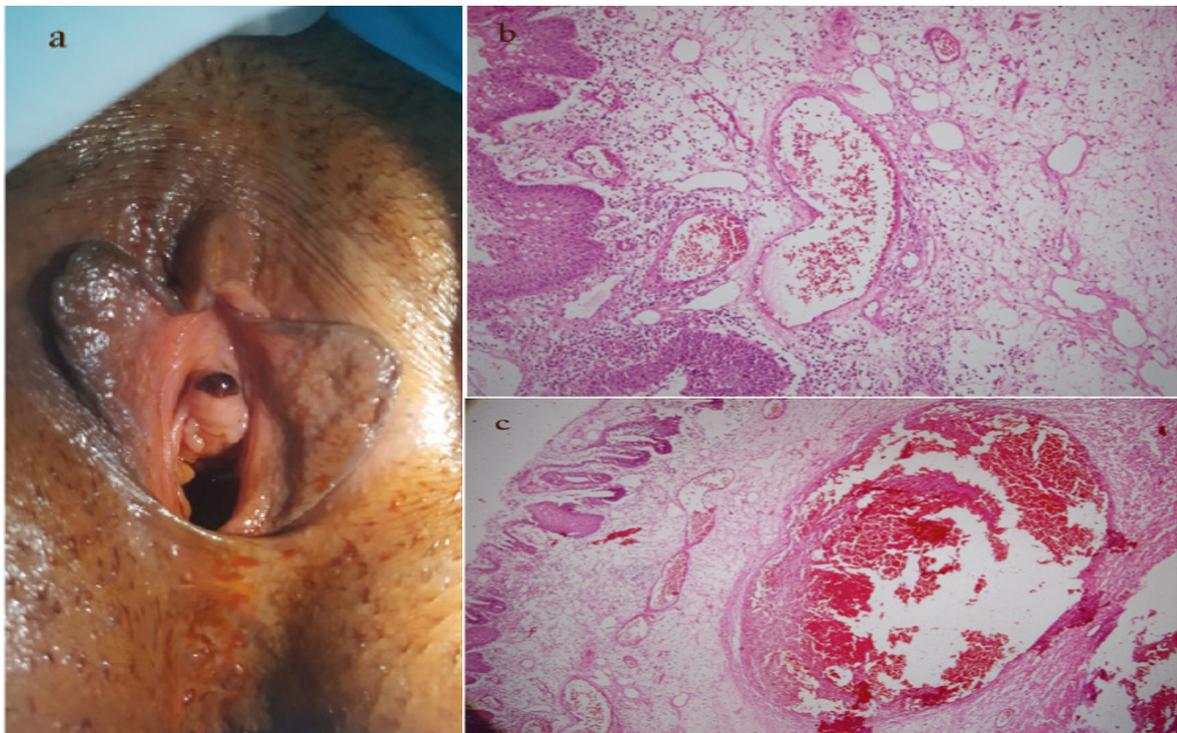
Genitourinary hemangiomas are very rare. To our knowledge few cases of female urethral hemangiomas have been reported and presenting cases are the first reports in Iran.

They should be considered as differential diagnosis of any female patient with microscopic or gross hematuria or bloody urethral discharge, especially when other parts of urinary system are radiologically intact. Thorough physical examination of genital area is highly recommended in order not to miss any urethral lesions. Herein we report two cases of female urethral cavernous hemangioma, their management and a review of literature.

**Keywords:** urethral hemangioma; hematuria; urethral mass; cavernous hemangioma; female urethral mass

### INTRODUCTION

Hemangioma of the urinary tract is an unusual entity and a few cases have been reported (Table 1). They may occur in all parts of the urinary tract but kidneys seem to be the most frequently affected followed by



**Figure 1. a:** 1x1 cm reddish urethral mass. **b&c:** Section show polypoid fragments of urothelial lined mucosa with presence of dilated cavernous vessels filled with blood and thrombus in lamina propria. Lymphocytic infiltration around vessels is prominent.

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**Table 1.** Reports of female urethral hemangioma in the literature.

Case	First Author	Journal	Year	Age of Patient
1	Uchida K.	J Urol.	2001	59
2	Tabibian L.	J Urol.	2003	81
3	Rao.A.R.	Urology	2005	26
4	Rohan VS.	Saudi J Kidney Dis Transpl.	2006	60
5	De Silva- Gutierrez A.	Rev Mex Urol.	2013	84
6	Ongun S.	Urol J.	2014	68
7	Bolat MS.	Pan Afr Med J.	2015	51
8	Regragui S.	Pan Afr Med J.	2016	61
9	Chiao-Ching Li.	Medicine	2017	8
10 (Present case1)	Sharifiaghdas F	Urol J.	2018	38
11 (present case2)	Sharifiaghdas F	Urol J.	2018	65

the bladder. They are generally seen as isolated lesions but they may be multiple and associated with congenital disorders such as klipple- Trenaunay syndrome , Sturge-Weber and systemic angiomasia<sup>(1)</sup>. Herein we report two cases of cavernous hemangioma of the urethra in female patients for the first time in Iran. To our best knowledge, there are less than ten documented case reports of female urethral cavernous hemangioma in the literature<sup>(2-7)</sup>.

## CASE REPORT

### Case one:

A 38 year old woman presented with painless urethral mass since 6 months ago. There was no complaint of gross hematuria or any other urinary symptoms. She mentioned episodes of mild bleeding from the mass in last few days. In physical examination a 1x1 cm round, reddish mass was seen in urethral meatus (**Figure 1, a**). The gross appearance was similar to a urethral caruncle. Urine analysis revealed microscopic hematuria. Ultrasound of genitourinary system and urine cytology were normal. In cysto-urethroscopic evaluation, the origin of the mass seemed to be in distal urethra. Other parts of

the urethra and urinary bladder were normal. Written informed consent was obtained from the patient and excisional biopsy of the mass was done at the same session. The mass had a broad base and was bigger than its initial view. The base of the mass was repaired with separate absorbable sutures. The pathology report revealed a cavernous hemangioma (**Figure1, b&c**).

### Case two:

A 65 year old female presented with a relatively large urethral mass since 20 days ago following constipation and straining. There was no dysuria, hematuria or other irritative urinary symptoms however she mentioned difficulty in voiding in last few days. In physical examination, a large peri-urethral mass with diffuse areas of thrombosis in its surface was noticed (**Figure 2, a**). In order to ameliorate obstructive urinary symptoms, a 16 French foley catheter was inserted in urethra and conservative management was initiated. Meanwhile, further evaluations including ultrasonography of genitourinary tract, urine analysis and urine cytology were performed. After 10 days of medical and conservative treatment including topical estrogen and oral antibiotic, the mass seemed to be smaller in size however it was



**Figure 2. a:** a large peri-urethral mass with diffuse areas of thrombosis in its surface. **b:** The same mass after conservative treatment.

not completely resolved (**Figure 2, b**). All para-clinical tests found to be normal except for microscopic hematuria. Cysto-urethroscopic evaluation under spinal anesthesia showed normal bladder but a mass originating from distal urethra. Excisional biopsy of the mass was performed at the same session using electro-cautery. In order to prevent subsequent meatal stenosis, eversion of urethral mucosa with simple 4-0 absorbable sutures was done properly. Foley catheter remained for another 7 days. The pathology report revealed a cavernous hemangioma with multiple foci of thrombosis. In one year follow up of two cases, neither of them had any complications or recurrence of the mass.

## DISCUSSION

The origin of hemangiomas is controversial but may arise from embryonic nests of unipotent angioblastic cells, which fail to develop into normal blood vessels<sup>(8)</sup>. The lesions are composed of a mixture of endothelium lined spaces containing erythrocytes and organized thrombi. They grow slowly and do not communicate with the surrounding vessels therefore are not truly invasive<sup>(1)</sup>. Mean age of presentation is 22 years, while age ranges from 3 to 68 years<sup>(3)</sup>. Urethral hemangiomas are the least common genitourinary hemangiomas<sup>(9)</sup>. Female urethral hemangiomas are very rare and ninety percent of urethral hemangiomas are seen in men. The most common symptom of urethral hemangiomas is hematuria either microscopic or gross<sup>(3)</sup>. Sometimes the bleeding can be very intense and cause anemia<sup>(6)</sup>. Other clinical presentations are: intermittent urethral bleeding and discharge, meatal mass, urinary retention, and lower urinary tract symptoms<sup>(3,9,10)</sup>. Differential diagnosis for female urethral mass includes: urethral caruncle, urethral prolapse, peri-urethral abscess, warts, urethral polyp, granuloma gravidarum, leiomyoma, malignancies like Squamous cell carcinoma, Transitional cell carcinoma, Sarcoma, and Melanoma<sup>(6)</sup>. Urethroscopy is the diagnostic method of choice and will define the site and extent of the lesions<sup>(3)</sup>. MRI is another useful diagnostic modality<sup>(2,4)</sup>. Accurate diagnosis requires pathologic confirmation. Treatment options depend on location and size of the mass and management should be individualized. Asymptomatic lesions do not require any treatment<sup>(1)</sup>. Hemangiomas are often underestimated. Even small lesions may extend further than is immediately obvious<sup>(1)</sup>.

Small isolated lesions are treated endoscopically by electrocoagulation, fulguration or Nd:YAG, Argon or KTP laser ablation<sup>(8,9)</sup>. Electro fulguration carries the risk of urethral scarring<sup>(1)</sup>. Other treatment options include: local resection, topical as well as oral steroids, sclerotherapy, and cryoablation<sup>(5,8,10)</sup>. In case of local recurrence after endoscopic ablation, open exploration and wide excision is recommended<sup>(1)</sup>.

Although urethral hemangiomas are very rare, they should be considered as differential diagnosis of any female patient with microscopic or gross hematuria or bloody urethral discharge. We believe that in "large masses" surgery is the treatment of choice and local medical treatment will change the gross appearance of the mass in a way that diagnosis would be very difficult with no benefit for the patient .

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