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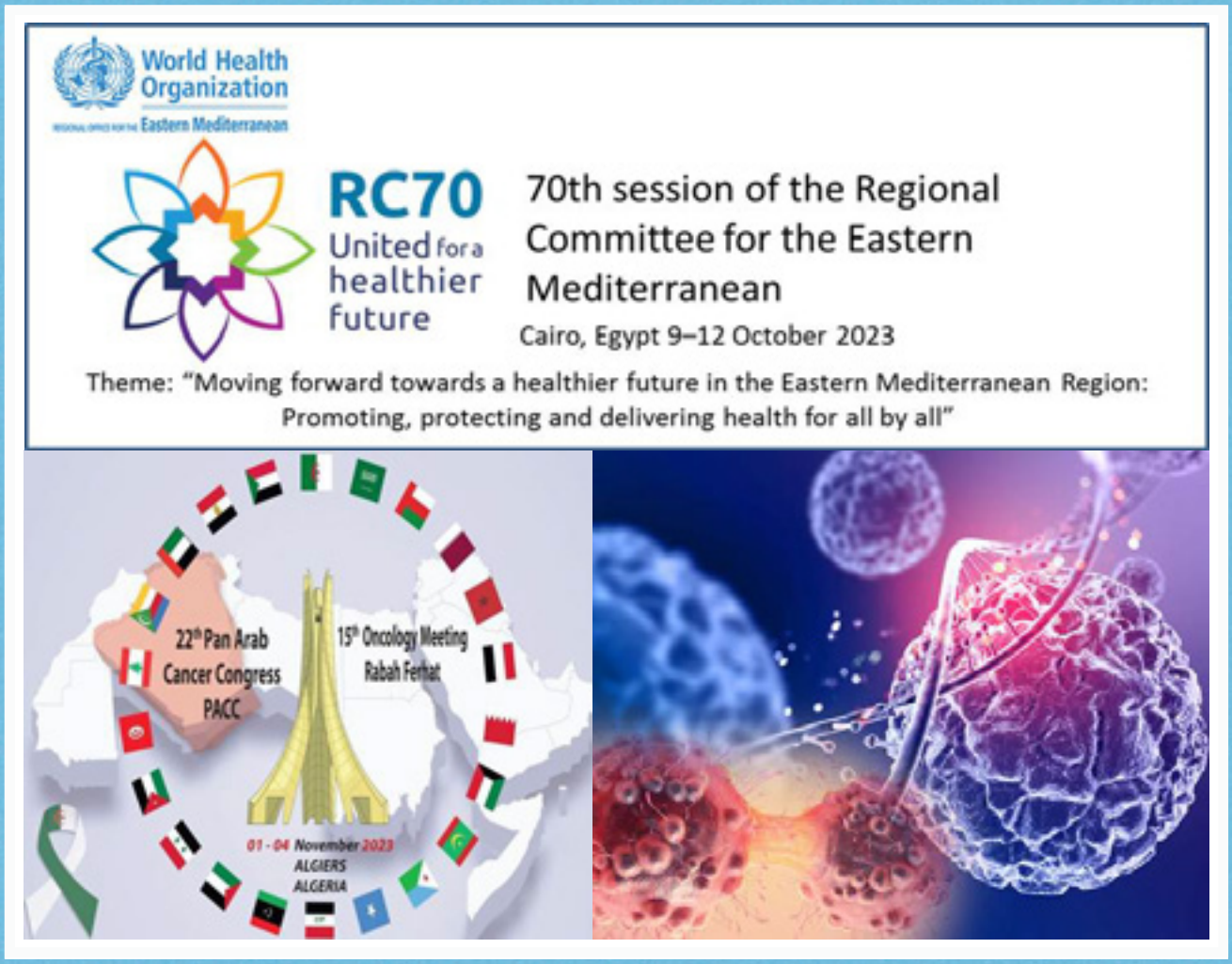

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Case Report

An Unusual Cause of Recurrent Visible Hematuria; Posterior Urethral Hemangioma: A Case Report and Review of Literature

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Abstract:

Urinary tract hemangiomas are uncommon entity to urologists, despite their benign nature and course; they can cause significant patient's hemodynamic instability or distress if not recognized and managed properly. Here, we present a case of urethral hemangioma with its treatment, follow–up course and review of literature of similar cases.

Introduction: Bloody urine or urethral bleeding can be the initial presentation of a number of different medical and surgical conditions, some of which are benign and others are malignant, when encountered, they are alarming and must be evaluated thoroughly. As a cause, urethral hematomas are faced rarely, among all urinary tract sites; they are the second least common[1]. Due to their paucity in clinical practice, there is no agreed approach on their management and each case has to be individualized. In the literature, different assessment and treatment methods were tried with satisfactory outcomes in each one. Here we report a case with detailed, concise explanation of such pathology and a comprehensive comparison in light of previous literature.

Keywords: Urethral hemangioma, urethral bleeding, benign urinary tract tumors

Case presentation:

A 35–year–old male, who was first referred to our institution a year before hospitalization as a case of urethral mass diagnosed cystoscopically while being evaluated for a complaint of persistent bleeding per urethra.

Upon assessment, patient reported that he has been complaining primarily of spontaneous occasional initial painless bleeding per urethral for almost a year associated with clots and there were no other urinary symptoms. On direct questioning no reported related history of infections, trauma or urological procedures. He is known to have primary infertility due to issue related to his female partner, otherwise; he has insignificant medical and surgical histories. Clinical examination revealed a well–looking adult male with central obesity, otherwise; general and genitalia examinations were unremarkable. Relevant work–up of complete blood count (CBC), renal function test (RFT), coagulation profile, urinalysis (UA), urine culture and cytology along with contrasted computerized tomography (CT) scan of the urinary tract were unremarkable.

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With a preoperative provisional diagnosis of urethral mass, patient admitted electively and underwent urethrocystoscopic evaluation revealing a fixed lesion located at verumontanum, grossly measuring approximately 1 cm and associated with surrounding hypervascular urethral mucosa (figure 1), bladder scanning

Literature	Presentation	Management	Follow–up
Qian et al. (2022)[3]	36–year–old male presented with recurrent postejaculatory hematuria. Clinically was stable. Urethrocystoscopy revealed single hemangiomatous lesion at 5 o'clock of posterior urethra.	Excision by plasma electrodes and urethral catheter removed after 2 days	No clinical recurrence
Genov et al. (2022)[4]	64–year–old male presented with recurrent intermittent urethral bleeding. Clinically was stable, Urethrocystoscopy revealed single hemangiomatous lesion at navicular fossa.	Coagulation by Thulium YAG (Tm:YAG) LASER and urethral catheter (size 18–French) removed after 1 day	No clinical recurrence
Masood et al. (2021)[5]	18–year–old male presented with recurrent intermittent urethral bleeding. Clinically was pale. Urethrocystoscopy revealed multiple hemangiomatous lesions at anterior urethra.	Fulguration by diathermy initially, then bleeding recurred which was managed by intralesional Triamcinolone and urethral catheter removed after 3 days	No clinical recurrence
Raquel et al. (2019)[6]	61–year–old male presented with recurrent urethral bleeding associated with obstructive lower urinary tract symptoms with history of previous urethral hemangioma excision. Clinically was stable. Urethrocystoscopy revealed multiple hemangiomatous lesions at anterior urethra and prostatic urethra with bulbar stricture	Visual internal urethrotomy for the stricture and conservative management for all hemangiomas and urethral catheter removed after 5 days	No clinical recurrence
Farzaneh et al. (2019)[7]	38–year–old female presented with urethral mass associated with bleeding. Clinically had a single palpable urethral mass. Urethrocystoscopy revealed an extension of the mass up to distal urethra.	Surgical excision	No clinical recurrence
Farzaneh et al. (2019)[7]	65–year–old female presented with urethral mass. Clinically had a single palpable urethral mass. Urethrocystoscopy revealed an extension of the mass up to distal urethra.	Excision by electrocautery and urethral catheter (size 16–French) removed after 7 days	No clinical recurrence
Khadijeh et al. (2018)[8]	70–year–year–old female presented with urethral bleeding associated with painful micturition. Clinically had a palpable urethral mass. Urethrocystoscopy revealed single dark urethral mass of few millimeters in size (Masson's hemangioma)	Surgical excision	No clinical recurrence
Mohammad et al. (2017)[9]	41–year–old male presented with painless visible hematuria and urethral bleeding. Clinically was stable. Urethrocystoscopy revealed single hemangiomatous lesion at anterior urethra.	Coagulation by diathermy	Presented four weeks later with recurrent urethral bleeding which was managed by Holmium YAG (Ho:YAG) LASER
Mohammad et al. (2017)[9]	22–year–old male presented with recurrent postejaculatory hematuria. Clinically was stable. Urethrocystoscopy revealed single hemangiomatous lesion at 12 o'clock of anterior urethra.	Coagulation by Holmium YAG (Ho:YAG) LASER and urethral catheter (size 18–French) removed after 2 days	No clinical recurrence
Souhail et al. (2016)[10]	61–year–old female presented with urethral bleeding associated with painful micturition. Clinically had a single urethral mass at 6 o'clock. Urethrocystoscopy revealed an extension of the mass up to proximal urethra.	Surgical excision and urethral catheter removed after 3 days	No clinical recurrence
Ioannis et al. (2009)[11]	27–year–old male presented with intermittent urethral bleeding. Clinically was stable. Urethrocystoscopy revealed single hemangiomatous lesion at anterior urethra.	Excision by biopsy forceps and urethral catheter (size 20–French) removed after 2 days	No clinical recurrence
Naoki et al. (2007)[12]	24–year–old male known case of Klippel–Trenaunay syndrome (KTS) presented with urethral bleeding. Clinically was stable. Urethrocystoscopy revealed multiple hemangiomatous lesions at anterior urethra.	Endoscopic sclerotherapy by injecting 5% solution of monoethanolamine oleate (Oldamine)	Slight bloodstained urethral discharge
Wilson et al. (2001)[13]	36–year–old male known case of Blue Rubber Bleb Nevus syndrome (BRBNS) presented with urethral bleeding. Clinically was stable. Urethrocystoscopy revealed single hemangiomatous lesion at anterior urethra.	Conservative, bleeding settled after 2 days	No clinical recurrence

Previously reported cases of urethral hemangiomas in adults

Literature	Presentation	Management	Follow–up
Pal et al. (2023)[14]	14–year–old male presented with recurrent visible hematuria with history of previous urethral hemangioma excision. Clinically was pale with low Hb level (6.6 g/dL). Urethrocystoscopy revealed two hemangiomatous lesions at 8 and 12 o'clock of anterior urethra.	Coagulation by Holmium YAG (Ho:YAG) LASER and urethral catheter removed after 5 days	No clinical recurrence
Mohammad et al. (2017)[9]	14–year–old male presented with painless urethral bleeding. Clinically was stable. Urethrocystoscopy revealed multiple hemangiomatous lesions at 4 o'clock of anterior urethra.	Coagulation by Holmium YAG (Ho:YAG) LASER and urethral catheter (size 16–French) removed after 1 day	Presented two weeks later with recurrent urethral bleeding which was managed by Holmium YAG (Ho:YAG) LASER
Abdulkadir et al. (2011)[15]	8–year–old male known case of Klippel–Trenaunay syndrome (KTS) presented with urethral bleeding. Clinically was stable. Urethrocystoscopy revealed single hemangiomatous lesion at anterior urethra.	Conservative, bleeding settled after urethral catheterization	No clinical recurrence

Previously reported cases of urethral hemangiomas in pediatrics

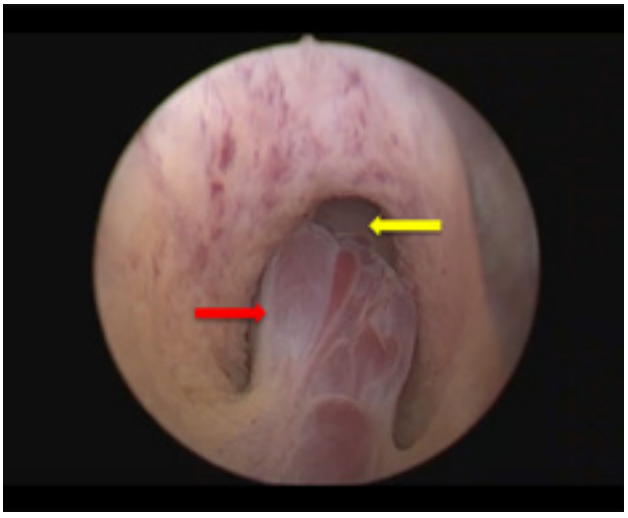

Figure 2. Post resection and fulguration

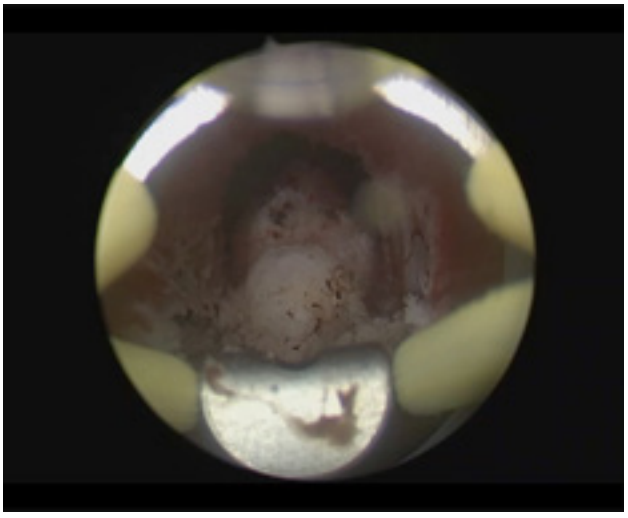

Figure 2. Post resection and fulguration

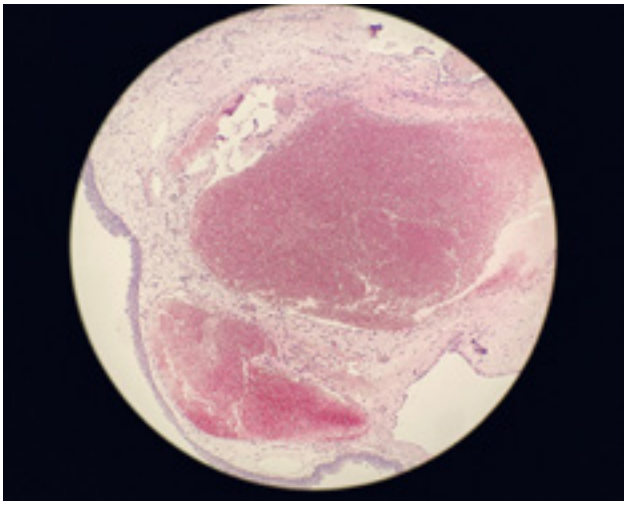

Figure 3. A well–circumscribed benign vascular tumor lined by urothelium, composed of blood vessels lined by single layer of flattened endothelial cells with no evidence of malignancy

was unremarkable, using a resectoscope sized 26–French the mass resected completely and sent for histopathological assessment in a Formalin container, hemostasis secured (figure 2), at the end; an open–tip urethral catheter sized 14–French inserted over a guidewire.

48–hours later, patient was asymptomatic and vitally stable, with no hematuria, catheter removed and patient discharged. On follow–up visits, patient reported no recurrence of primary complaint or other urinary concerns. Histopathological evaluation came back as benign tumor composed of dilated blood vessels with an outer surface lined by normal urothelium consistent with a hemangioma (figure 3).

Discussion:

Hemangioma is a vascular anomaly characterized by abnormal growth of a blood vessel cells, those vascular anomalies are classified into either malformations or tumors, the latter are sub classified into benign, locally aggressive and malignant, hemangiomas are considered benign tumors[2]. On a molecular level, they tend to grow in a budding–like manner, pushing but not invading the surrounding tissue, histologically composing of multiple spaces that are lined by endothelium and filled by erythrocytes with or without the presence of thrombi[1].

Urinary tract hemangiomas are infrequent; they tend to occur as solitary pathology or less commonly as part of congenital disorders such as Blue Rubber Bleb Nevus syndrome (BRBNS) and Klippel–Trenaunay–Weber syndrome (KTWS). Urethral hemangiomas in particular are fairly rare, only a few small series and case reports in the literature have been reported. For unknown reason, urethral hemangiomas have a male predominance reaching to a ratio of 9:1 in comparison with females, most present in the second or third decade of life, they may occur as single or multiple lesions that can be located anywhere along the urethra, but it has been observed that the posterior urethra, specifically the area between the verumontanum and external sphincter is the commonest location[3].

The clinical manifestation of urethral hemangiomas is dependent on the extent and location of the hemangioma. Anatomically and histologically, the male phallus has limited tissue comprising both cavernous bodies, those are half–encapsulated at their distal end and fully encapsulated along their penile shaft part to the base of the phallus, because of this; they are usually asymptomatic. When a hemangioma grows toward the urethra they become symptomatic. In males, they manifest in descending order as intermittent visible hematuria, spontaneous bloody urethral discharge, hematospermia or postejaculatory hematuria, they are typically painless unless if associated with thrombosis. Sexual activity, trauma and infections are the main contributing factors for the clinical appearance; this may explain their prevalence among young adults. Urethral mass is the main presenting feature in females[3].

A number of adjunct diagnostics may be considered based on the primary presenting complaint, relevant associated history and clinical assessment. Because the commonest presenting symptom is visible hematuria, ultrasonography and contrasted CT of urinary tract are usually conducted. When such condition is suspected, magnetic resonance imaging (MRI) is to be considered preoperatively to help guide surgical intervention if intended. The definitive diagnosis is made by cystoscopy and biopsy, they grossly may appear as red or bluish sessile polypoid or flat lesion surrounded by dilated blood vessels (varicosities) with histopathology exhibiting cavernous or capillary types with the former being commoner[1].

Management of urethral hemangiomas is case–based, taking into consideration the clinical presentation, lesion characteristics and patient's bothersome. After careful assessment, they can be treated conservatively, medically or surgically; all modalities have been reported with nearly similar outcomes (Table 1 and 2). Patients, who are asymptomatic or not bothered, functionally good, have small lesions and are stable clinically maybe observed; given that those lesions might fade spontaneously which is one of the natural courses of hemangiomas[1].

Variety of medical therapies are being used, their principal effect is to stop the growth and induce regression of the lesion, possibly by inhibiting angiogenesis and inducing apoptosis, they are delivered orally or locally through direct injection into the lesion. Oral Propranolol, a non–selective beta–blocker, narrows the abnormal hemangioma vessels due to its vasoconstrictive effect stopping their growth and proliferation which will lead to apoptosis, due to its immediate effect; it is consider now first–line medical therapy, but it has been less used due to patients being bothered by its side effect profile. Intralesional corticosteroids namely Triamcinolone have been used with a good results in the literature, its mechanism of action is not fully understood but it is theorized that it has an inhibitory effect on the hemangioma stem cells through inhibition of vascular endothelial growth factor A (VEGF–A) synthesis, in the past; it was the first–line medical option but currently many will prefer to avoid using steroids due to its wide range of mild to serious side effects. Another option is the use of anti–tumors, primarily Pingyangmycin; it induces DNA chain breakage, production of oxygen free radicals interfering with cell cycle and division ultimately leading to apoptosis.

The most widely practiced way of treatment of urethral hemangiomas nowadays is surgical intervention; those are highly effective with excellent outcome and prognosis. One general principal must be remembered is that those hemangiomas often recur locally so ensuring total removal with negative margins and clear base is to be complemented. Different modalities have been described in the literature and choosing between them is dependent on multiple factors including overall patient's status, presentation and degree of bothersome, number, location and character of the hemangioma, lastly is availability of instruments. Primary excision, fulguration and coagulation via LASER or plasma electrodes have yield the most successful outcomes of no clinical recurrence postoperatively in comparison with sclerotherapy that was associated with mild postoperative mild bloodstained urethral discharge which subsided weeks later. In females, the commonest clinical presentation is urethral mass that can be noted on physical examination, special consideration here is to assess the proximal extent of the mass by cystoscopy to determine the proper curative intervention that usually is total surgical excision.

Complications are uncommon, this is maybe because of both the benign nature of urethral hemangiomas and the simple surgical principals applied, since there is urethral intervention, urethra–related symptoms and acquired disorders mainly urethral stricture is to be expected.

Depending on the selected method, one can expect immediate bleeding, urinary retention due to blood clots, urinary incontinence due to external urethral sphincter injury, erectile dysfunction due to nerve injury, impotence due to obstructive prostatic ducts, difficulty voiding due to urethral stricture and non–specific lower urinary tract symptoms.

After the primary intervention has been successfully attempted, urethral catheter is usually fixed in place for tamponading effect and to allow for initial healing. Currently, there is no agreed statement on which size and how long before it should be removed, on both age groups; adults and pediatrics who received different surgical intervention, the timeframe ranged between 1 to 7 days with the primary indication for removal being clear urine output. Observation for spontaneous clear micturition before discharge is advised. Because of the favorable postoperative course of urethral hemangiomas; follow up does not require further invasive measures. Regular clinical assessment focusing on urethral symptoms, signs and associated possible complications is adequate.

In our case, patient demonstrated the typical physical behavior of such benign tumor of intermittent painless visible hematuria, which was located at the posterior urethra, complete excision and fulguration intended with the expected favorable postoperative course of no clinical recurrence.

Conclusion:

This article represent the second reported case of posterior urethral hemangioma in the gulf region that responded well to surgical management. Urethral hemangiomas are rare and benign, and it should be included in the differential diagnosis of urethral bleeding, hematuria or hematospermia. They are usually asymptomatic until they grow towards the urethra or become subjected to increased urethral pressure, thus recognizing them maybe challenging. Although imaging techniques might be helpful in identifying the tumor, it is difficult to make a definitive diagnosis before cystoscopic evaluation and histopathological examination. The decision to employ active therapy must be determined by the individual features of the hemangioma. In our case, the rarity of such pathology makes it of a value and similar cases should be reported worldwide.

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Conflict of interest:

None declared

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