

Periurethral Hemangioma - A Rare Case

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ABSTRACT

Hemangiomas are benign vascular tumors, rarely seen in the genitourinary tract. A 45-year old man, with a history of urethral trauma and varicocele surgery was admitted to urology department with hematuria and hard urination. Cystourethroscopy revealed 3 cm urethral stricture on the proximal urethra and urethroplasty was applied. Microscopically, periurethral fibrovascular tissue, fibrosis in striated muscle, thickening on the walls of vessels, anastomosing irregular splits and cavernous hemangiomas forming papillary structures were revealed. Immunohistochemically the lining cells of vascular walls are CD 31 (+)-used primarily to demonstrate the presence of endothelial cells, D2-40 (-)-selectively expressed in lymphatic endothelium. We reported the lesion as 'periurethral hemangioma'. After treatment the patient is symptom-free without any episodes of urethral bleeding. In literature only little more than 20 cases of periurethral hemangiomas have been reported. Here, we want to share this rare lesion, which can cause hematuria, hematospermia and obstructive symptoms.

Key words: periurethral, hemangioma, genitourinary tract

PERİÜRETRAL HEMANJİYOM - NADİR BİR VAKA

ÖZET

Hemanjiyomlar, nadir vasküler tümörler olup, genitouriner sistemde nadiren bulunurlar. 45 yaşında üretral travma ve varikosel operasyonu olan hasta, hematüri ve işeme güçlüğü şikayeti ile üroloji kliniğimize başvurdu. Sistoskopisinde proksimal üretrada 3 cm'lik darlık izlendi ve üreteroplasti uygulandı. Mikroskopik olarak periüretral fibrovasküler doku, çizgili kasta fibrozis, damar duvarlarında kalınlaşma, birbiriyle anastomozlaşan yarıklar ve kavernöz hemanjiyomatöz yapılar oluşturan papiller alanlar izlendi. Vasküler duvarı döşeyen hücreler; primer olarak endotelial hücrelerce ekspres edilen CD 31 ile immunreaktivite gösterirken, lenfatik epitelde ekspres edilen D2-40 ile immunreaktivite izlenmedi. Lezyonu 'periüretral hemanjiyom' olarak raporladık. Tedavi sonrasında hastada üretral kanama olmamış ve semptomsuz olarak hayatına devam etmektedir. Literatürde 20'nin üzerinde periüretral hemanjiyom olgusu bildirilmiştir. Biz de bu nadir görülen; hematüri, hematospermi ve obstrüktif semptomlara yol açabilen lezyonu paylaşmak istedik.

Anahtar sözcükler: periüretral, hemanjiyom, genital trakt

Hemangiomas are benign vascular tumors. The most involved sites are liver and skin. Genitourinary tract is an uncommon site for hemangiomas. In literature only a little more than 20 cases of urethral hemangiomas have been reported (1). Urethral hemangioma commonly presents as urethral bleeding, obstructive symptoms, hematuria and/or hematospermia. Their origin is still controversial (2). Treatment depends upon site, size and number of hemangiomas (3).

Here, we present a case of urethral hemangioma which is an extremely rare clinical condition.

Case report

A 45-year-old man admitted to urology department with hematuria and hard urination. He had a history of urethral trauma and varicocele operation. Cystourethroscopy revealed 3 cm urethral stricture on the proximal urethra and urethroplasty was applied. Periurethral tissue was sent to our pathology department. Macroscopically, it was four fragmented reddish-brown fragile tissue. On histological examination periurethral fibrovascular tissue, fibrosis in striated muscle, thickening on the walls of vessels, anastomosing irregular splits and cavernous hemangiomas forming papillary structures were revealed (Figure 1). Immunohistochemically the lining cells of vascular walls

are CD 31(+)-used primarily to demonstrate the presence of endothelial cells (Figure 2), D2-40(-)-selectively expressed in lymphatic endothelium (Figure 3). As a result we reported it as periurethral hemangioma. After treatment the patient is symptom-free without any episode of urethral bleeding.

Discussion

Hemangiomas of the urinary tract are rare. They localise in the kidney, ureter, bladder, prostate and urethra. Urethral hemangiomas are also rare. It can occur at any age, generally men are effected. Only little more than 20 cases have been reported in literature (1). It can be associated with the presence of cutaneous hemangiomas and Klippel-Trenaunay syndrome (4). The most common type is cavernous hemangioma. Urethral hemangioma commonly presents as urethral bleeding, hematuria and/or hematospermia (5). Large lesions can cause obstructive urinary symptoms or protrude through the urethral meatus (6).

Hemangiomas can regress spontaneously as a result of fibrosclerosis. If asymptomatic, they do not require treatment (1). If symptomatic, treatment depends upon site, size and number of hemangiomas (3). Treatment ranges from transurethral approach to open reconstructive surgery (7).

Urethral hemangiomas have varying size and histologically they consist of thin walled vascular spaces lined by endothelial cells (7).

The case presented herein was a 45-year old man who had hematuria, hard urination and urethral stricture in cystourethrosopy. Urethroplasty was applied. On histological examination anastomosing irregular splits and cavernous hemangiomatous forming papillary structures were revealed. To make differential diagnosis with lymphangioma, immunohistochemistry was applied. The lining cells of vascular walls are CD 31(+)-used primarily to demonstrate the presence of endothelial cells, D2-40 (-)-selectively expressed in lymphatic endothelium. As a result we reported it as periurethral hemangioma. After treatment the patient is symptom-free without any episode of urethral bleeding.

The origin of periurethral hemangiomas is still controversial. One suggestion about their origination is differentiation from unipotent angioblastic cells that fail to develop into normal blood vessels (2). Local varicosity and chronic

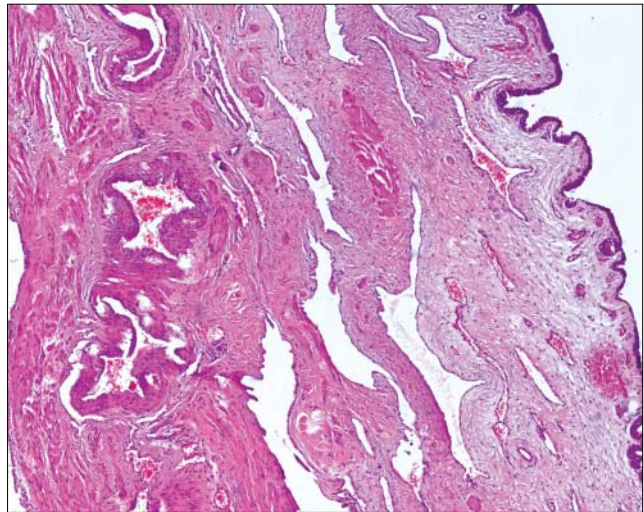


Figure 1. Anastomosing irregular splits and cavernous hemangiomatous forming papillary structures (H-Ex40)

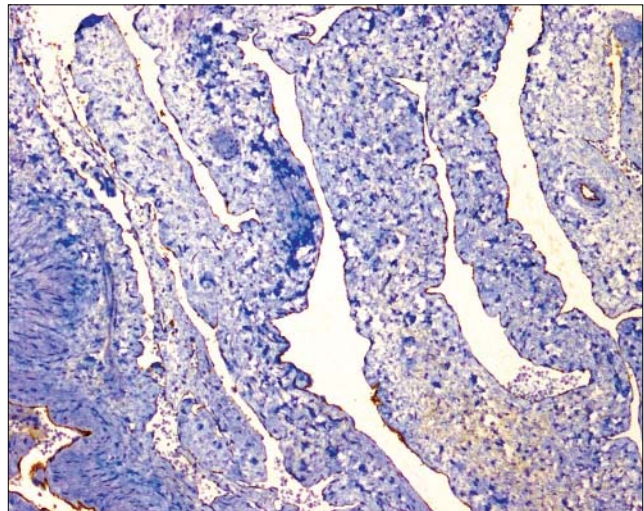


Figure 2. CD 31(+) endothelial cells (x100)

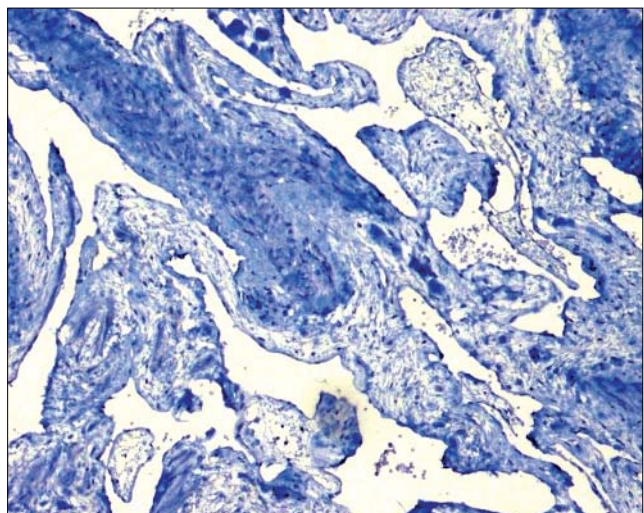


Figure 3. D2-40(-) endothelial cells (x100)

irritation were also suggested as etiologic factors (8). The latter theory has newly supported by Alfonsa et al. who presented a 86-year old woman with urethral hemangioma (9). Our patient had a history of urethral trauma and varicosel operation. At that point our patient also support the second theory. He also had also chronic nonspecific inflammation on microscopic examination. During the

follow-up period of 20 months he had neither recurrence nor any urologic complaints.

Consequently, urethral hemangiomas are rare benign vascular tumors. Urologists should keep in mind this for differential diagnosis when a patient apply with obstructive urinary symptoms, bleeding, hematuria and/or hematospermia.

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