CLINICAL CASE

Urethral hemangioma: a case report

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KEYWORDS
Hemangioma, urethral tumor, urethrorrhagia, Mexico.

Abstract Hemangiomas are benign vascular tumors and their presence at the level of the urethra is extremely rare. Incidence is higher in men and there are very few reports of cases in women. We present herein the case of an 84-year-old woman that presented with urethral bleeding that often led to an anemic syndrome requiring blood transfusions. Physical examination showed a 2 x 2 cm tumor at the level of the meatus and the external third of the urethra (urethral hemangioma). We carried out a surgical resection of the tumor as well as a percutaneous cystostomy, complementing its management with 800cGy of local radiotherapy, and in this way stopping the bleeding. According to published reports, hemangiomas are secondary to angioblast cell degeneration of embryonic remnants. We believe that chronic inflammatory, as well as atrophic, degenerative changes, could play an important role as a coadjuvant factor in the formation of these tumors in the advanced age patient.
Introduction

Hemangiomas are benign tumors of vascular origin that can present during any decade of life. The most commonly affected organs are the skin and liver. They can develop at all levels of the urinary system, including the kidney, ureter, bladder, prostate, and urethra. Their presence in the latter is perhaps less frequent; there is a greater incidence in men, and reports in women are extremely rare. Tumors can be single or multiple and can present in any segment of the urethra. The cardinal sign is the presence of bleeding, meaning either gross or microscopic hematuria, urethrorrhagia, or hematospermia.

Case presentation

An 86-year-old woman had a past medical history of breast cancer treated with radical mastectomy, radiotherapy, and adjuvant chemotherapy 16 years prior. She currently presented with dilated cardiopathy and controlled high blood pressure. Illness onset was one month before, characterized by intermittent monosymptomatic hematuria and urethrorrhagia, which increased in frequency and intensity one week prior to hospital admittance, resulting in an anemic syndrome that required a blood transfusion in order to be corrected. Physical examination revealed a vascularized tumor that was dependent on the inferior commissure of the urethral meatus of approximately 2 x 2 cm (Figure 1). Urinary tract ultrasound sonography (USG) image identified no alterations, and urethrocystoscopy showed that the tumor extended to the first third of the urethral floor. It bled easily and the cystoscope was able to pass to the posterior urethra, the bladder neck, and the bladder, showing no apparent alteration. Careful observation revealed that both ureteral meatuses ejaculated clear urine.

Resection of the tumor was carried out (Figures 2 and 3). The histopathologic study reported a urethral hemangioma with large dilated venous vessels between a lax stroma, as well as ureteritis cystica with Von Brunn’s nests. Immunohistochemistry CD34 and alpha actin for endothelium and smooth muscle showed an irregular profile of vascular proliferation; smooth muscle cells did not mark CD34 and did not express estrogen and testosterone hormone receptors (Figures 4 and 5). At one month from the surgical procedure the patient presented with severe urethrorrhagia, and so a second surgery was performed in which there was a wider resection that included the middle third of the urethra, additional fulguration of the surgical site, and percutaneous cystostomy as urinary diversion. After a new episode of hematuria, even though it was mild, 15 days later the patient was given local external direct electron beam radiotherapy with two sessions of 400cGy, achieving definitive remission of the bleeding at the 12th month of follow-up.

Discussion

Hemangiomas generally are single tumors but they may form part of the Sturge Weber and Klippel-Trenaunay-Weber syndromes. In relation to etiology and pathogeny, the most accepted theory describes an angioblast degeneration of embryonic remains that ends by forming these tumors of mesenchymal origin. However, it cannot be ruled out that they could also be due to degenerative processes associated with chronic and atrophic irritative factors, such as in the present case, that developed in a patient over 80 years of age.

Clinical urethral hemangiomas can be asymptomatic or present with episodes of monosymptomatic hematuria,
Urethrorrhagia, or hematospermia of great intensity and clotting, causing the patients to develop an anemic syndrome requiring blood transfusion, as was the case with our patient.

Urethrocystoscopy is an excellent diagnostic method that allows for the identification of the characteristics, friability, size, location, and number of hemangiomas; it also enables the planning of possible therapeutic options for each particular case.3,6

Differential diagnosis of a urethral hemangioma must be made with urethral caruncles, polyps, leiomyomas, periurethral abscesses, and malignant tumors such as adenocarcinomas, transitional cell tumors, sarcomas, melanomas, and squamous cell carcinomas.

Treatment consists of tumor resection or ablation. It should be taken into account that hemangiomas can deceptively be larger than they clinically appear to be, due to possible very deep implantation in the internal layers of the urethra, which could make complete extirpation difficult in one surgery and cause new episodes of bleeding, requiring repeat treatments, as in our patient.

Different treatment modalities have been described, such as: transurethral resection and fulguration of the base of the tumor, laser ablation, cryotherapy, selective embolization, steroid use, local radiotherapy, and open surgical excision.2,4,7,9 In some cases, urethrectomy and urinary diversion are also necessary.10 As long as the resection is complete, outcome is favorable.

In our case, complete extirpation was not possible in just one surgical resection due to the important infiltration of the tumor in the lamina propria of the urethra that could not be macroscopically identified. The patient’s age and
general status were not conducive to a wider resection or radical urethrectomy with a continent urinary diversion, and thus local radiotherapy became the most viable option, offering definitive control of the tumor. Percutaneous cystostomy was very helpful, because it avoided irritating factors at the surgical site and resolved the problem of diaper management of micturition in an elderly patient with limited mobility, keeping her dry.

This type of tumor should always be kept in mind in the diagnosis of patients presenting with recurrent monosymptomatic hematuria, urethrorrhagia, or hematospermia.

Finally, as in all aspects of our medical practice, the general conditions of each case in particular should be taken into account when contemplating therapy proposals, so that the best one is chosen.

Conflict of Interest

The authors declare that there is no conflict of interest.

Financial disclosure

No financial support was received in relation to this article.

References