Urology Case Reports 23 (2019) 52–54

Contents lists available at ScienceDirect
Urology Case Reports

journal homepage: www.elsevier.com/locate/eucr

Trauma and reconstruction

Urethral cavernous hemangioma in female: Surgical resection and reconstruction with ventral vaginal graft urethroplasty

Kadek Budi Santosa, Pande Made Wisnu Tirtayasa*, I.Wayan Yudiana, Gede Wirya Kusuma Duarsa, Anak Agung Gde Oka
Division of Urology, Department of Surgery, Faculty of Medicine, Universitas Udayana/Sanglah Hospital, Bali, Indonesia

ARTICLE INFO

Keywords:
Urethral hemangioma
Female
Graft
Urethroplasty

ABSTRACT

Urinary hemangiomas are uncommon and can predispose any part of the urinary system. The urethra is infrequently involved and usually affected male urethra. We reported a sixty-three-year-old female patient with intermittent urethral bleeding due to tumor arising at the edge of the urethra. Urethrocystoscopy was done prior to surgical resection and ventral vaginal graft urethroplasty was performed directly afterwards. Histopathological reported a urethral cavernous hemangioma. An individualized approach regarding the most appropriate procedure for a given patient should be recommended.

Introduction

Cavernous hemangioma is a rare benign vascular tumor. Some studies have reported cavernous hemangioma in the female urethra and most of them were located at distal third of the urethra. The tumor usually found superficial and most of the studies have reported good results after a simple surgical resection.1–3 However, none of them clearly stated the technique and difficulties found during tumor resection.

The surgical technique can be challenging if the tumor base is wide and many urethral tissues must be removed to prevent recurrence. In this presentation, we report a case of urethral cavernous hemangioma in female where a ventral vaginal graft urethroplasty was performed directly after extensive urethral tumor resection. To our knowledge, only a few cases have been reported and this is the first documented case of female urethral hemangioma from Indonesia.

Case presentation

A 63-year old female patient presented with intermittent urethral bleeding. She also reported poor urinary stream, nocturia up to four times a night and a sensation of incomplete emptying. Physical examination showed a vascularized tumor arising at the edge of the urethra (Fig. 1). Urethrocystoscopy confirmed that the tumor involved almost half length of the urethra and the tumor base arise from ventral part of the urethra.

Tumor resection was carried out extensively using an 11-blade scalpel. The incision was performed carefully following the margin of the tumor and deeply through periurethral bed tissue. Almost half lumen of the entire urethral wall at the ventral side was resected in advance to have a complete tumor removal (Fig. 2). Vaginal mucosa was harvested in a standard fashion from the patient’s right lateral vagina. The graft’s donor defect was closed with a 4-0 vicryl running suture. A ventral inlay technique was used to place the graft to the urethral defect. Interrupted 4-0 vicryl sutures were placed to circumferentially anastomose the graft to the edge of the defect. A 14-Fr urethral Foley catheter was inserted and was maintained for 5 days. Pathological examination showed cavernous vascular spaces filled with blood and separated by connective tissue stroma revealed as cavernous hemangioma of the urethra (Fig. 3).

The patient was symptom-free at 3 months of follow-up with no evidence of tumor recurrence, no stress incontinence, and no need for urethral dilation. In addition, uroflowmetry test showed a good result.

Discussion

Hemangioma is a rare benign vascular tumor that can occur at any age, commonly found on the skin and liver. However, hemangioma can grow on urinary tract, including kidney, ureter, bladder, prostate, and urethra. They usually congenital and develop from the embryonic rest of the unipotent angioblastic cells that fail to develop into normal blood vessels.1 The most common clinical manifestation are hematuria and
obstructive symptoms. Diagnosis is primarily confirmed by the lesion’s histological analysis. Differential diagnosis includes formation resulting from urothelial reactions, such as inflammatory processes and, in post-menopausal women, urethral caruncles, and urethral polyp. Urethrocystoscopy is an excellent diagnostic method that supports in identification of the characteristic, fragility, size, location, and number of hemangiomas.2,3

The management of female urethral hemangioma can be challenging due to the chance for urethral stricture. Despite benign nature, hemangioma may recur due to incomplete excision. In our case, the choice was performing ventral vaginal graft urethroplasty since the tumor base was wide and extended to the ventral urethral tissue which must be resected. Primary closure will likely make the urethral lumen narrower. Vaginal mucosa graft was chosen because it was practically available, hairless elastic surface, good vascularity, and early healing.4

We did not find stress incontinence in our patient after 3 months of follow-up. Ventral resection may have a theoretically benefit in this matter due to the intact pubo-urethral ligament and the striated muscle was relatively deficient posteriorly. Some studies showed that stress urinary incontinence was rare after urethroplasty for female urethral syndrome (FUS), it remains unknown whether the dorsal or ventral urethral resection would prove a preferential approach from the standpoint of disruption of the external urethral sphincter.5

Surgeons should continue to innovate and search for the most effective techniques, while aiming to minimize patient morbidity and potential complications in managing urethral hemangioma in female patients.

Conclusion

Surgical resection followed by direct reconstruction with vaginal mucosa graft urethroplasty is an effective choice for the treatment of cavernous hemangioma urethral in female. Moreover, an individualized approach tailoring the most appropriate procedure for a given patient should be advocated.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.eucr.2018.12.008.

References

1. Ahuja A, Sen AK, Bhardwaj M. Cavernous hemangioma of anterior urethra: an unusual

Fig. 1. Clinical appearance of cavernous urethral hemangioma.

Fig. 2. Urethral hemangioma was excised.

Fig. 3. Histopathology view of the specimen revealed a urethral cavernous hemangioma (H&E x10).
cause of vaginal bleeding. *Indian J Pathol Microbiol.* 2016;59:245–246.

2. Bolat MS, Yuzuncu K, Akdeniz E, Demirdoven AN. Urethral cavernous hemangioma in a female patient: a rare entity. *Pan Afr Med J.* 2015;22:352.

3. Ongun S, Celik S, Aslan G, Yorukoglu K, Esen A. Cavernous hemangioma of the female urethra: a rare case report. *Urol J.* 2014;11(2):1521–1523.

4. Hoag N, Gani J, Chee J. Vaginal-sparing ventral buccal mucosal graft urethroplasty for female urethral stricture: a novel modification of surgical technique. *Investig Clin Urol* 2016;57(4):298–302.

5. Haderer JM, Pannu HK, Genadry R, Hutchins GM. Controversies in female urethral anatomy and their significance for understanding urinary continence: observations and literature review. *Int Urogynecol J Pelvic Floor Dysfunct.* 2002;13(4):236–252.