

A case of severe bladder wall hypertrophy: bladder cancer or sequela of bladder outlet obstruction?

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Angiosarcomas are soft tissue malignancies of connective tissue origin with rapid hematogenous spread, but are extremely uncommon primary tumors of the bladder with approximately ten reported cases in the 20th century. We report a 59-year-old man with benign prostatic hyperplasia (BPH) and gross hematuria who underwent a bladder biopsy for a markedly thickened bladder wall on CT scan. Biopsy specimens demonstrated deep vascular

malformations that were concerning for a malignancy. Intense pathological review was initially not definitive and transurethral resection of the prostate (TURP) was performed for his symptoms. Interestingly, surgery reversed the bladder process. This case serves as a unique example of how the sequelae of bladder outlet obstruction (BOO) can resemble a malignant process on presentation but represent no more than hypertrophic adaptations in the bladder wall musculature of a patient with BPH and significant outlet obstruction.

Key Words: benign prostatic hyperplasia, bladder wall hypertrophy, angiosarcoma

Case report

A 59-year-old man presented with a 3 day history of hematuria and dysuria. His urological history was significant for benign prostatic hyperplasia (BPH), lower urinary tract symptoms (LUTS) treated with

finasteride and terazosin, and an elevated serum prostate specific antigen 7 years prior that prompted prostate biopsies. Biopsies were negative, but a 98 cc gland was identified.

On acute presentation, the large prostate was noted and urine cytology revealed atypical urothelial cells. Computed tomography (CT) showed diffuse, marked bladder wall thickening and pericyclic stranding and inflammatory changes consistent with an active infectious/inflammatory or neoplastic process, Figure 1a.

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On cystoscopy, an extremely irregular bladder wall mucosa with friability, edema and irritation was noted. The biopsies showed a polypoid cystitis and papillary hyperplasia but underlying these epithelial changes was an extensive infiltrate of varying sized, congested vessels with reactive endothelium and some hob nailing, Figure 2a. Deep in the detrusor muscle was a network of irregular, dilated channels with muscular walls, Figures 2b and 2c. The initial differential diagnosis ranged from benign vascular malformation to angiosarcoma. Consultation was sought for a unifying diagnosis of both the superficial and deep vascular changes and to exclude malignancy. In the end, no unifying diagnosis was possible, and the final diagnosis was superficial granulation tissue and deep vascular malformation.

At 1 month follow up, the patient returned with LUTS. Transurethral resection of the prostate (TURP) was completed without complications and 52 g of tissue were resected. At 3 months follow-up, CT scan showed markedly reduced bladder wall

thickness, Figure 1b, attributable to the surgical relief of obstruction.

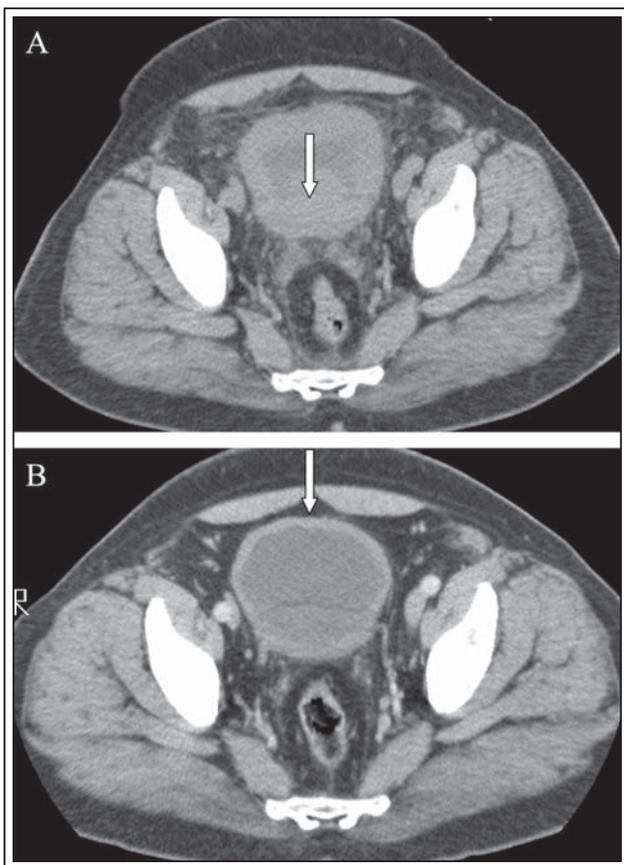


Figure 1. Helical CT scans showing bladder wall thickness before (panel A) and 3 months following TURP (panel B).

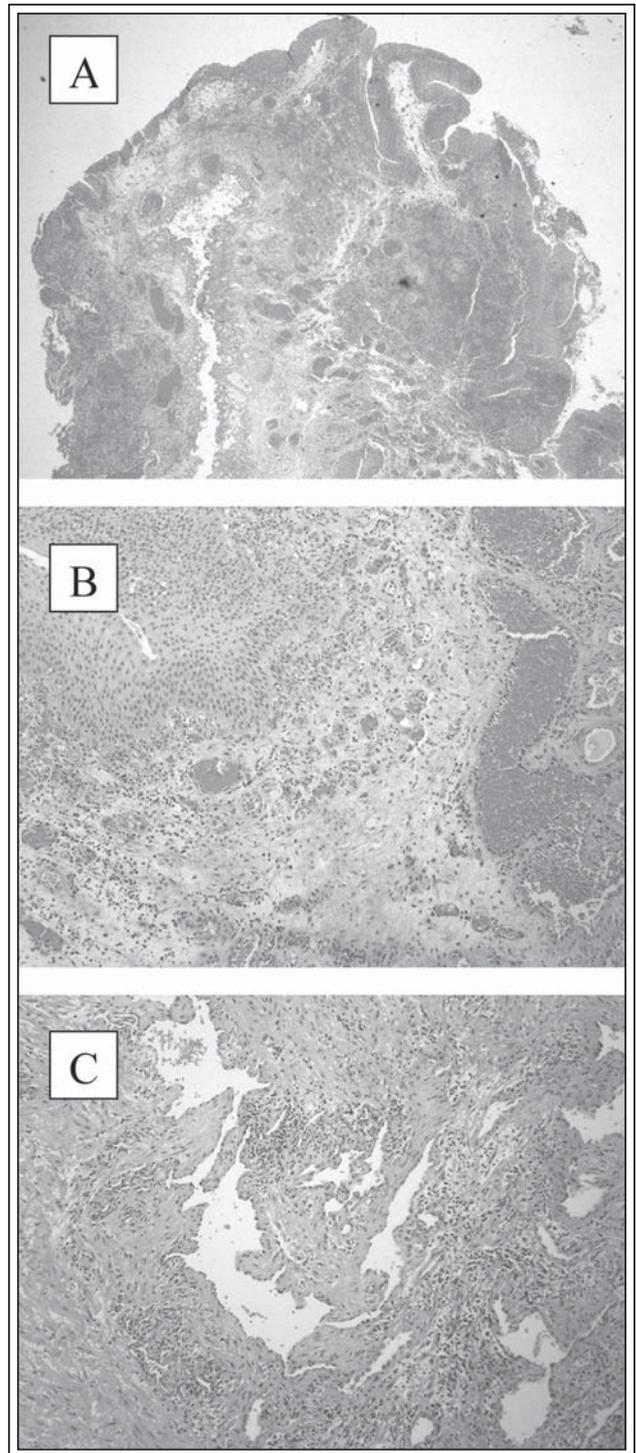


Figure 2. Panel A, superficial bladder wall with numerous and congested vessels. Panel B, variably sized superficial vessels and reactive endothelium. Panel C, deep, dilated vascular channels.

Discussion

Hematuria can result from a wide variety of underlying disease processes affecting the bladder, ureters, and kidneys. Although many cases of microhematuria are transient and benign, persistent macrohematuria is a common clinical feature of bladder cancer, especially in men over the age of 50.¹ Bladder wall thickening, as seen in Figure 1a, was thought to represent an active neoplastic process in this patient, and while the histopathological findings could not definitively exclude a primary bladder malignancy like an angiosarcoma, the clinical improvement of the patient following surgical resection of the prostate suggested the process was benign.

Angiosarcomas are soft tissue malignancies of connective tissue origin with rapid hematogenous spread. They make up less than 2% of all malignancies and are extremely uncommon tumors of the bladder with approximately 12 reported cases in the literature.²⁻¹³ The most common presenting symptom of a primary bladder angiosarcoma is hematuria, but dysuria, suprapubic or groin pain, bladder outlet obstruction and weight loss have also been reported. There is a strong male predilection with a male to female ratio reported to be 8:1. On cystoscopic evaluation, raised edematous lesions involving the trigone is a common finding, but the bladder dome, diverticula and ureteral orifice have all been involved as well. Characteristic histopathological findings include poorly differentiated vascular malformations coupled with papillary endothelial cell proliferation. Regardless of tissue of origin, the overall 5 year survival rate for angiosarcomas is less than 35%.¹⁴ However, one group recently reported a case of 6 year survival in a man with primary bladder angiosarcoma following a radical cystectomy and adjuvant chemotherapy and radiation.¹⁵ No study, however, has specifically evaluated the best treatment strategy for patients diagnosed with primary bladder angiosarcomas and given the exceedingly low prevalence of this disease, this study would be difficult to implement.

In this case, the bladder wall hypertrophy is attributable to the urethral obstruction by an enlarged prostate.¹⁶ Bladder outlet obstruction has been previously shown in animal models to induce adaptations in the bladder wall that include smooth muscle cell hypertrophy, decreased detrusor muscle contractility, and formation of trabeculated epithelium.^{17,18} Interestingly, bladder wall hypertrophy has been shown to be one adaptation that is reversible when the obstruction is surgically relieved. Although the pathophysiological details of this relationship is not completely understood, this case

serves as a unique example of the clinical manifestations observed in this phenomenon as well as illustrates how this clinical presentation can be remarkably similar to a more malignant process, such as bladder cancer. □

References

1. Messing WM. Urothelial Tumors of the Bladder. Wein AJ, Kavoussi LR, Novick AC, Partin AW, Peters CA: *Cambell-Walsh Urology* 9th edition, volume 3, Section XV, Chapter 75 2445.
2. Engel JD, Kuzel TM, Moceanu MC et al. Angiosarcoma of the bladder: a review. *Urology* 1998;52:778-784.
3. Jungano F. Sur un cas d'angio-sarcome de la vessie. *Annales des maladies des organes genitourinary*. 1907;25:1451-1461.
4. Casal J, Singer ED, Monserrat JM. Angiosarcoma of bladder. *Rev Argent Urol Nefrol* 1970;39:53-55.
5. Wasmer Jm, Block NL, Politano VA et al. Penile angiosarcoma presenting in bladder. *Urology* 1981;18:179-180.
6. Schwartz RA, Kardashian JF, Mcnutt NS et al. Cutaneous angiosarcoma resembling anaplastic Kaposi's sarcoma in a homosexual man. *Cancer* 1983;51:721-726.
7. Nanus DM, Kelsen D, Clark DC. Radiation-induced angiosarcoma. *Cancer* 1987;60:777-779.
8. Stroup RM, Change YC. Angiosarcoma of the bladder: a case report. *J Urol* 1987;137:984-985.
9. Morgan MA, Moutrous DM, Pippitt CH JF et al. Vaginal and bladder angiosarcoma after therapeutic irradiation. *South Med J* 1989;82:1434-1436.
10. Aragona F, Ostardo E, Prayer-Gayletti T et al. Angiosarcoma of the bladder: a case report with regard to histologic and immunohistochemical findings. *Eur Urol* 1991;20:161-163.
11. Ravi R. Primary angiosarcoma of the urinary bladder. *Arch Esp Urol* 1993;46:351-353.
12. Schindler S, De Frias DV, Yu GH. Primary angiosarcoma of the bladder: cytomorphology and differential diagnosis. *Cytopathology* 1999;10:137-143.
13. William S, Romaguera R, Kava B. Angiosarcoma of the bladder: case report and review of the literature. *Scientific World Journal: TSW Urology* 2008;8:508-511.
14. Mark R, Poen J, Tran L et al. Angiosarcoma: a report of 67 patients and a review of the literature. *Cancer* 1996;77:2400-2406.
15. Pazona JF, Gupta R, Wysock J et al. Angiosarcoma of bladder: long-term survival after multimodal therapy. *Urology* 2007;69:575.e9-575.e10.
16. Kojima M, Inui E, Ochiai A, Naya Y, Kamoi K et al. Reversible change of bladder hypertrophy due to benign prostatic hyperplasia after surgical relief of obstruction. *J Urol* 1997;158:89-93.
17. Andersson KE. Storage and voiding symptoms: pathophysiologic aspects. *Urology* 2003;62:3-10.
18. Gosling JA, Gilpin SA, Dixon JS et al. The effect of age on the autonomic innervation of the urinary bladder. *J Urol* 1986;136:501-504.