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Vasculitis of the bladder: An extremely rare case report

Radwan Kassir^{a,*}, Pascal Mouracade^b, Gabriele Barabino^a, Michel Peoc'h^c,
Muriel Cuilleron^d, Marc Gigante^b^a Department of General Surgery, CHU Hospital, Jean Monnet University, Saint Etienne, France^b Department of Urology, CHU Hospital, Jean Monnet University, Saint Etienne, France^c Department of Pathology, CHU Hospital, Jean Monnet University, Saint Etienne, France^d Department of Radiology, CHU Hospital, Jean Monnet University, Saint Etienne, France

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ABSTRACT

INTRODUCTION: Isolated vasculitis of the bladder is extremely rare. The main causes of which are auto-immune diseases and occasionally infections. Corticosteroid therapy plays a central role in treatment in the majority of cases.**PRESENTATION OF CASE:** We report a case of gross hematuria associated with irritative low urinary tract symptoms (LUTS) and an increase of biological parameters of inflammation. Radiologic studies suspected a pelvic tumor process. We performed a cystoscopy with multiple biopsies. The pathological findings of the chips were in favor of a thrombotic nongranulomatous vasculitis of small and medium caliber. In view of these findings, all systemic diseases and inflammatory diseases such as cryoglobulinemia, the anti-phospholipid syndrome, Crohn's disease were eliminated. The symptoms regressed completely under antibiotics and anticoagulants.**DISCUSSION:** Our treatment options were based on the extent of the acute phase reaction and the pelvic venous thrombosis. A few similar cases have been reported in the literature, particularly a case of isolated necrotizing vasculitis of the bladder involving small vessels with a mild laboratory acute phase reaction which was treated with corticosteroids and cyclophosphamide.**CONCLUSION:** It is important to differentiate this rare pathological feature of the bladder from other bladder tumors as the treatment is medical rather than surgical.© 2013 Surgical Associates Ltd. Published by Elsevier Ltd. Open access under [CC BY-NC-ND license](http://creativecommons.org/licenses/by-nc-nd/4.0/).

1. Introduction

The vasculitides are inflammatory diseases of the blood vessel wall, the main causes of which are auto-immune diseases and occasionally infections. Corticosteroid therapy plays a central role in treatment in the majority of cases. The current classification distinguishes the various vasculitides according to the types of vessels involved. Whereas most vasculitides are rare diseases they generally involve various groups of organs, only rarely including the bladder. We report a case of isolated vasculitis of the bladder presenting with gross hematuria and an acute phase reaction in a young person.

2. Presentation of case

Mr. S., a 31 year-old man, presented to the emergency department in mid-January with hypogastric pain present for 48 h.

* Corresponding author at: Department of General Surgery, CHU Hospital, Jean Monnet University, Avenue Albert Raimond, 42270 Saint Etienne, France.
Tel.: +33 6 13591971; fax: +33 4 77127015.

E-mail address: Radwankassir42@hotmail.fr (R. Kassir).

He had no personal or family medical or surgical (including urological) history and had no history of medical drug use. The patient was a smoker of 5 packet years and occasionally drank alcohol. He was a cabinet-maker by occupation.

Clinical history revealed bilateral lumbar pain with sudden onset gross hematuria and fever (38.2 °C). He described burning on micturition and urgency and also gave a history of bloody diarrhea which resolved without treatment over the weeks before this episode.

On physical examination he had bilateral lumbar tenderness and more diffuse abdominal pain. A rectal examination was normal and painless with no melena or rectal bleeding. Clinical examination was otherwise unremarkable.

Laboratory tests on admission showed leukocytosis of $27 \times 10^9/L$ (leukocytes $20 \times 10^9/L$) with a CRP of 186 mg/L, creatinine of 84 $\mu\text{mol/L}$, normal coagulation profile and an increased fibrinogen of 7.5 g/L. His urine cytology and blood cultures were subsequently negative.

An urgent contrast enhanced abdominal and pelvic CT scan showed asymmetrical bladder wall thickening with contiguous pelvic thromboses and also infiltration of the pelvic floor fat fusing beneath the peritoneum with no renal lesions or dilatation of the ureteric, pyelo-caliceal system. Because of the pelvic thrombosis

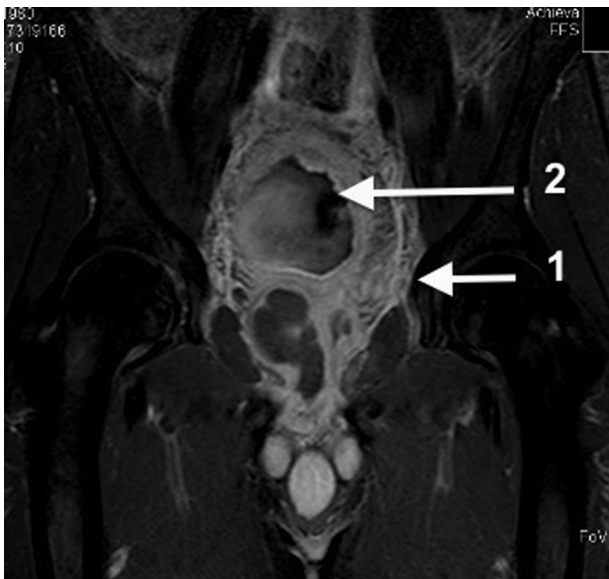


Fig. 1. Abdominal and pelvic MRI. 1, pelvic varicose veins; 2, circumferential thickening of the bladder wall.

we also performed a Doppler lower limb ultrasound which showed no superficial or deep vein thrombosis.

Abdominal and pelvic MRI (Figs. 1 and 2) showed large pelvic varices with circumferential thickening of the bladder wall predominantly on the left lateral aspect, between 16 and 18 mm thick. There were two possible differential diagnoses after this initial assessment, either purely inflammatory bladder disease with multiple pelvic thromboses or a neoplastic process with a botryoid bladder sarcoma.

The patient was started on dual antibiotic therapy with ofloxacin/gentamicin (because of his leukocytosis, pending the results of the various samples) and anticoagulation (because of the pelvic venous thrombosis).

A transurethral resection of his bladder (TURB) was performed on D8 after admission. Macroscopically this showed a tumor on the left lateral aspect of the dome of the bladder which was infiltrating in appearance. Because of the extent of the lesion the resection was incomplete and the fragments were sent to the histology laboratory.

Effective anticoagulation was impossible because of the extent and persistence of his hematuria despite the TURB and a caval filter was inserted.

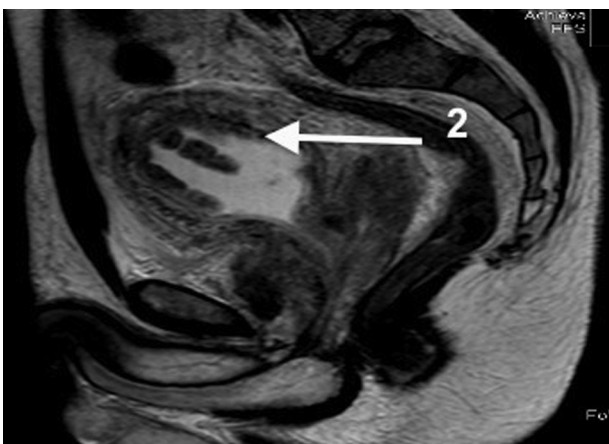


Fig. 2. Abdominal and pelvic MRI. 1, pelvic varicose veins; 2, circumferential thickening of the bladder wall.

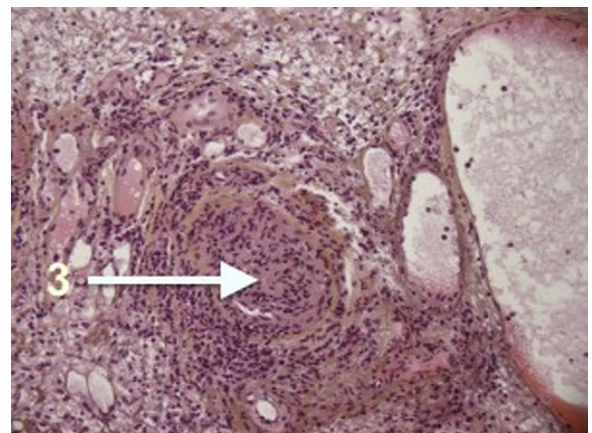


Fig. 3. Histology of the fragments; 3, thrombosed veins.

A chest-abdominal and pelvic scan was requested on D10 after admission. He had a few lateral aortic lymph nodes under a centimeter in size in his abdomen with infiltration of the pelvic floor and bladder wall thickening with bulky creeping pelvic appearances compatible with venous thrombosis. No thoracic abnormalities were present.

Histological results from the fragments (Figs. 3 and 4) concluded that there were no signs of malignancy or eosinophilic infiltration: “histological appearances of non-granulomatous, small and medium diameter vessel thrombotic vasculitis”. An internal medicine opinion was requested after the histological result was received.

His thrombosis screen was completely negative and colonoscopy and gastroscopy were entirely normal. We did, however, find very profound iron deficiency with strongly positive ASCA antibodies and decided to request a videocapsule and enteric MR. Both of these investigations were normal.

The patient had no further hematuria during his urological follow up and gave no history of losing weight. A repeat CT scan found no further bladder thickening or pelvic thrombosis and cystoscopy was normal. Overall the patient had improved 9 months after the acute episode and no cause for his presentation was found.

3. Discussion

We have reported a case of vasculitis of the bladder complicated by pelvic deep vein thrombosis in a young person. The clinical

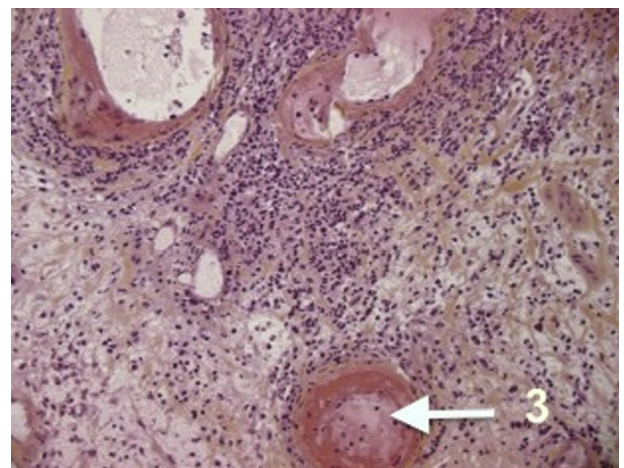


Fig. 4. Histology of the fragments; 3, thrombosed veins.

picture resolved entirely with anticoagulation and dual antibiotic therapy. Our treatment options were based on the extent of the acute phase reaction and the pelvic venous thrombosis. A few similar cases have been reported in the literature, particularly a case of isolated necrotizing vasculitis of the bladder involving small vessels with a mild laboratory acute phase reaction which was treated with corticosteroids and cyclophosphamide. This treatment was effective, the episode resolving over a few weeks.¹ Another case of vasculitis of the bladder without pelvic thrombosis has been reported, with complete remission on IV steroids.²

No cause was found in this case although this is not always the case. Wegener's granulomatosis³ and HBV infection⁴ have been implicated in some cases reported in the literature. It is important to differentiate this rare pathological feature of the bladder from other bladder tumors as the treatment is medical rather than surgical.

Conflict of interest statement

None.

Funding

None.

Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contributions

Radwan Kassir: writing; Michel Peoc'h: data collections; Pascal Mouracade: data collections; Gabriele Barabino: data collection; Muriel Cuilleron: data collections; Marc Gigante: study design.

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