



## A rare presentation of eosinophilic cystitis in paediatric urology

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### ABSTRACT

Eosinophilic cystitis (EC) is a rare inflammatory condition of the urinary bladder. It is extremely rare in childhood and may present with haematuria, lower abdominal mass or recurrent urinary tract infections. We present the case of a 5-year-old girl with recurrent, painless visible haematuria. Ultrasound showed a bladder mass and left hydronephrosis. Cystoscopy and bladder biopsy confirmed the diagnosis. The patient was successfully managed with oral treatment. In the case of EC, cystoscopy and bladder biopsy is key for diagnosis. Conservative management of unilateral hydronephrosis in EC is appropriate and we advise oral corticosteroids in its treatment.

### 1. Introduction

Eosinophilic cystitis (EC) is extremely rare in children, with just over 100 cases reported until this date.<sup>1,2</sup> It was first described by Brown in the 1960s as a form of granuloma of the urinary bladder.<sup>3</sup> Its presenting symptoms include lower urinary tract symptoms (LUTS), dysuria and visible haematuria.<sup>2</sup> The differential diagnosis may include interstitial cystitis, carcinoma in situ (CIS) and bladder cancer.<sup>2</sup>

### 2. Case presentation

A 5-year-old girl presented with an episode of visible haematuria for 1 week. She was brought by her parents to our urology clinic and her mother mentioned a similar episode that occurred a month before. Detailed history revealed the presence of visible haematuria, no dysuria, no LUTS or abdominal pain. She has no known allergies and no history of skin rashes, itching or wheezing. Her past medical history was insignificant.

The general examination was normal and her abdominal examination revealed no tenderness with no palpable abdominal mass. Ultrasound evaluation of the kidneys and urinary bladder showed left moderate hydronephrosis and a thickened urinary bladder wall with multiple mass-like lesions, mainly seen on the left lateral wall [Fig. 1]. Urinalysis showed only red and white blood cells.

Suspicion of a urinary bladder tumour was high, and the patient was referred for further diagnostic imaging. Focused ultrasound examination

reported an abnormal, asymmetrical hypoechoic urinary bladder wall thickening, indicating possible urinary bladder lymphoma. Computerised tomography (CT) scan with contrast confirmed the findings of a urinary bladder mass and left hydronephrosis. Intravenous pyelography and a micturating cystogram, to exclude vesicoureteric reflux, showed left hydronephrosis with no reflux and a filling defect in the urinary bladder. Unfortunately, bladder rupture occurred and extravasation of contrast was noticed at the end of the cystogram [Fig. 2]. This was managed conservatively with a urethral catheter left in-situ for 2 days until the haematuria resolved. Suspicion of lymphoma was still high, and the patient was scheduled for cystoscopy and urinary bladder biopsy one week after the catheter was removed.

The patient's parents were fully informed of what was suspected and the need for biopsies for histopathological examination. The operation was performed in a specialist paediatric clinic where appropriate sized paediatric endourology instruments were available. Cystoscopy showed a hyperaemic mucosa, mainly at the trigone, with nodules and multiple tumour-like structures in the left lateral urinary bladder wall. The left ureteric orifice appeared to be involved by these oedematous lesions. The right lateral wall showed a small area of ulceration with haemorrhagic spots. Cold biopsies were taken from the oedematous regions, right and left lateral bladder walls, dome and trigone of the urinary bladder and sent for histopathology examination. Urinary bladder capacity was normal, and a urethral catheter was inserted at the end of the procedure. The patient recovered well from the procedure and catheter removed on day 1 post-operation with clear urine. The patient was

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discharged on oral antibiotics and for follow up after histopathology results.

Histopathology specimens were sent to a specialised pathology lab and the report confirmed the presence of abundant eosinophils in the urinary bladder mucosa and lamina propria [Fig. 3]. These findings confirm the diagnosis of EC. The patient was referred to a paediatric consultant regarding further investigations and treatment options. Further investigations included serum Immunoglobulin A (IgA), which was significantly high (90 mg/dL) and a peripheral blood film that did not reveal any marked eosinophilia.

Treatment was started with oral prednisolone, antibiotics and anti-histamines. Regular follow up of treatment was maintained with the paediatrician and a follow up urology visit was scheduled after 4 weeks. The first follow up visit for the patient revealed normal micturition with no dysuria or bothersome LUTS. Urine colour was normal with no episodes of haematuria and no suprapubic pain. Ultrasound examination showed complete resolution of left hydronephrosis, normal urinary bladder wall with no thickening or mass-like lesions. Patient responded to oral steroid treatment very well and the dose tapered over two weeks. The patient made a full recovery within 6 weeks of treatment and is scheduled for follow up within 6 months.

### 3. Discussion

EC can be a self-limiting disease.<sup>2</sup> However, about 30% may show a recurrence of the disease,<sup>1</sup> necessitating adequate follow up. Reported complications of eosinophilic cystitis include hydronephrosis and rarely urinary bladder rupture.<sup>4</sup> Diagnosing the disease requires cystoscopy and urinary bladder biopsy for histopathological evaluation.<sup>2</sup> Treatment options include systemic corticosteroids, antibiotics, and in some cases resection or open surgery.<sup>1,2</sup>

Specific guidelines in paediatric urology for managing eosinophilic cystitis is currently lacking.<sup>5</sup> Our case report of a 5-year-old girl with eosinophilic cystitis, presenting with recurrent visible haematuria, urinary bladder mass and unilateral hydronephrosis can be challenging. This case report highlights the importance of suspecting EC in paediatric patients. Although it is an extremely rare condition, it must be included in the differential diagnosis of children presenting with haematuria and a urinary bladder mass. The initial presentation of painless visible haematuria with a urinary bladder mass in our reported case was an alarming symptom. Usually the first differential diagnosis is of a sinister aetiology that mandates further evaluation. Our differential diagnosis included lymphoma and urinary bladder rhabdomyosarcoma. Use of further imaging, mainly a CT scan with contrast, is useful to exclude other pathology. Cystogram evaluation initially showed a filling defect. However, this has caused bladder rupture with extravasation, possibly



Fig. 2. Cystogram showing extravasation in the AP (top) and Oblique (bottom) views.

due to the effect of EC on bladder wall tissue and has been reported in association with rapid bladder filling.<sup>4</sup>

### 4. Conclusion

Cystoscopy and bladder biopsy is the only method to confirm the diagnosis of EC. We advise that no intervention for unilateral hydronephrosis is required in case of confirmed EC and it must be monitored with ultrasound to confirm resolution. We aim to highlight the clinical

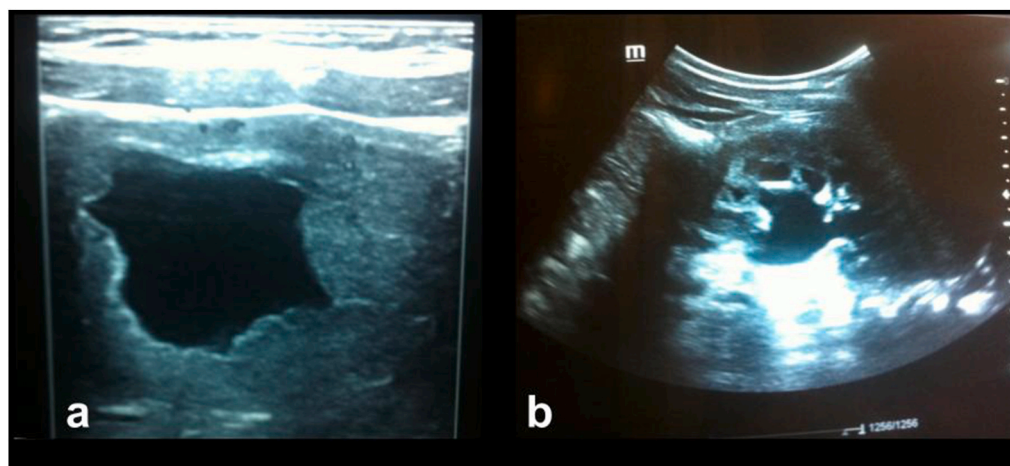


Fig. 1. Ultrasound images. (a) Thick wall of urinary bladder, with mass-like lesions. (b) Left moderate hydronephrosis.

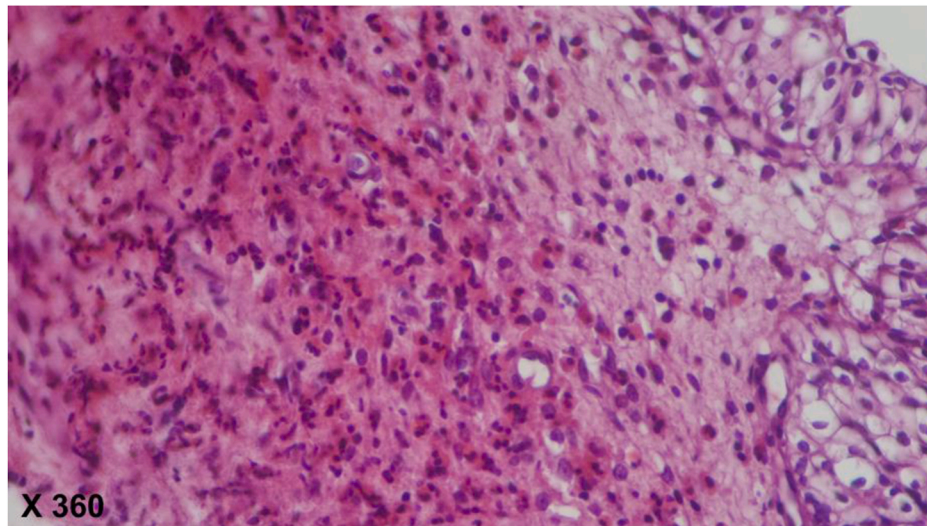


Fig. 3. Histopathology image showing abundant eosinophils infiltrating the mucosa and lamina propria.

significance of suspecting this rare disease in children, and our experience with managing such clinical presentation. We hope to expand the knowledge and management options for future cases, with an interest to advance guidelines for diagnosing and treating suspected cases of EC in paediatric urology.

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#### Ethical approval

Ethics approval not required.

#### Consent

Informed consent has been provided by the patient's legal guardian for this case report and accompanying images with guarantee of confidentiality.

#### Declaration of competing interest

There are no conflicts of interest, including specific financial

interests or relationships and affiliations relevant to the subject matter or materials discussed in the manuscript to be disclosed by any of the authors.

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