Journal of Pediatric Urology (2013) 9, 738—744

Journal of Pediatric urology

REVIEW ARTICLE

Eosinophilic cystitis in the pediatric population: A case series and review of the literature

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Received 5 July 2012; accepted 6 November 2012
Available online 4 February 2013

KEYWORDS
Eosinophilic cystitis;
Pediatric;
Inflammation;
Bladder

Abstract *Purpose:* Eosinophilic cystitis is a rarely seen condition in the pediatric population with indistinct symptomatology and non-standardized treatment protocols. We review our experience of treating patients with this condition.
Materials and methods: We retrospectively reviewed the records of four patients from a single institution who have been diagnosed and treated for eosinophilic cystitis. In addition, the literature was reviewed for cases of pediatric eosinophilic cystitis. Our patients were added and compared to this cohort.
Results: Our patients included 3 females and 1 male who range in age from 5 days to 18 years (5 days, 1 month, 7 years, 18 years). Both of the infants presented with a suprapubic mass and bilateral hydroureteronephrosis. The two older patients both had dysuria while the 18 yo also complained of fatigue, flank pain, and hematuria. Only 2 of the 4 patients were found to have significant peripheral eosinophilia and only one patient had eosinophiluria. All of the patients were diagnosed via cystourethroscopy with biopsy. Treatment in each case consisted of a combination of steroids, antihistamines, and antibiotics.
Conclusions: The presentation of eosinophilic cystitis is varied and diagnosis requires a high index of suspicion. Cystourethroscopy with biopsy is essential to establish the diagnosis as there is no typical appearance of the lesions or presenting signs/symptoms. Most cases of

Abbreviations: VUR, vesicoureteral reflux; CIC, clean intermittent catheterization; CGD, chronic granulomatous disease; EC, eosinophilic cystitis; WBC, white blood cells.

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http://dx.doi.org/10.1016/j.jpurol.2012.11.004

eosinophilic cystitis are responsive to medical therapy although in some cases recurrence may be noted.

Introduction

Eosinophilic cystitis is an inflammatory condition that has been well documented in adults. It has been described to a lesser degree in the pediatric population. Differences in the disease course and demographics in the pediatric versus the adult population make the diagnosis at times a quandary. Although presenting symptoms may be primarily irritative in nature, this form of cystitis is also difficult to diagnose without cystoscopy and subsequent bladder biopsy. Cystoscopy may reveal areas of mild erythema to gross bullous masses. Biopsy specimens will have prolific amounts of eosinophils in the tissue. To date, 52 pediatric patients have been reported in the literature, most as part of small case series. We review our practice's experience with the clinical diagnosis and treatment of eosinophilic cystitis and combine this with the previous 52 cases.

Case #1

A 5 day old female infant was admitted to the hospital due to pustules on her right upper extremity and neck. She was also noted to have a palpable suprapubic mass. Laboratory evaluation revealed peripheral eosinophilia with WBC of 8100 with 14% eosinophils. Urinalysis was notable for microscopic hematuria (5-10 RBC's/HPF) and minimal pyuria (0-2 WBC's/HPF). Urine, blood, and CSF cultures were all no growth. Culture of the pustules revealed *Staphylococcus aureus* and umbilical culture grew *Escherichia coli*, Klebsiella, and Proteus rettgeri.

Appropriate antibiotic therapy was initiated and an indwelling catheter was placed in the bladder resulting in only partial resolution of the suprapubic mass. A voiding cystourethrogram was performed which revealed multiple filling defects in the bladder wall. An intravenous pyelogram demonstrated bilateral hydronephrosis despite the indwelling catheter. Cystoscopy revealed diffuse yellow to red raised lesions, 5–10 mm in diameter over the entire mucosal surface of the bladder. Ureteral orifices could not be identified. Bladder biopsy showed an intact mucosa with marked vascular congestion (Fig. 1A) and eosinophilic infiltration of the submucosa and muscularis (Fig. 1B).

The biopsy findings prompted the addition of prednisone to the patient's treatment regimen. Repeat intravenous pyelogram, eleven days after the cystoscopy, revealed resolution of the hydroureteronephrosis.

The catheter was subsequently removed and the patient voided to completion without difficulty. The sulfa drug and steroids were continued for 4 more days and the infant was discharged to home. She was lost to follow up after discharge.

Case #2

A 7 year old male presented with a 6 month history of intermittent dysuria. Multiple urinalyses were normal. His past medical history was significant for chronic urticaria of unknown etiology for which he was taking daily cetirizine. He was also treated with a course of oral steroids for exacerbation of his urticaria. Physical exam on presentation was unremarkable with no focal abnormal findings.

Laboratory studies revealed a white blood cell count of 6700 with 1.6% eosinophils. An ultrasound demonstrated a mass in the anterior wall of the bladder. This finding was confirmed on a triphasic CT scan with the lesion measuring 2.1 × 3.3 × 2.3 cm (Fig. 2). No hydronephrosis or lymphadenopathy was identified. A cystoscopy revealed a yellowish mass protruding into the lumen from the left anterior wall of the bladder. Biopsies indicated an inflammatory lesion with bland appearing urothelial cells, a prominent submucosal layer expanded by mild fibrosis, and a background of severe inflammation dominated by eosinophils.

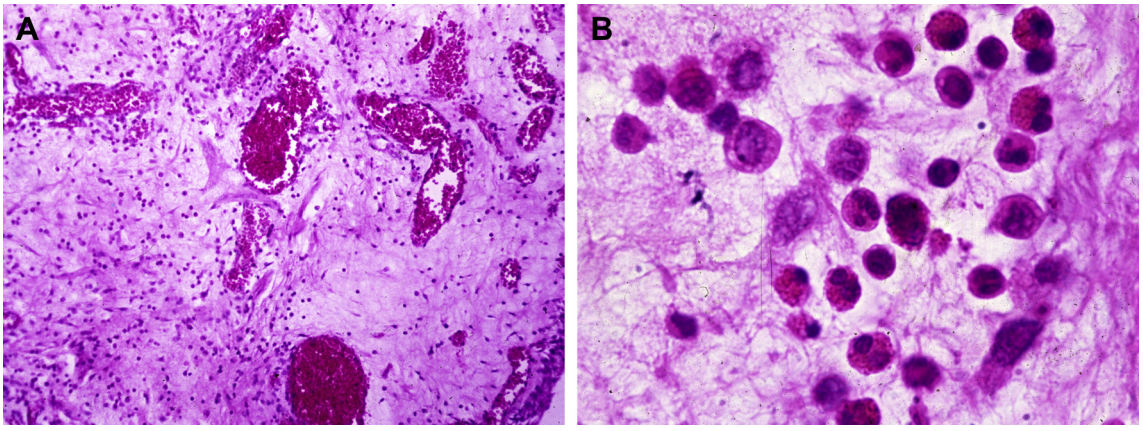

Figure 1 Bladder biopsy of patient #1 showed an intact mucosa with marked vascular congestion (A) and eosinophilic infiltration of the submucosa and muscularis (B).