REPORTE DE CASO

Cistitis eosinofílica en niños: un reporte de caso

Eosinophilic cystitis in children: a case report / cistites eosinofílica em crianças: um reporte de caso

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RESUMEN

La cistitis eosinofílica es una entidad poco común que se caracteriza por la inflamación con infiltrado eosinofílico de la pared vesical. No es muy frecuente en la edad pediátrica con mínimos reportes de casos en la literatura. Presentamos un caso de un paciente de 24 meses, a quien se le diagnostica dicha patología, el manejo instaurado y una revisión de la literatura.

Palabras clave: eosinofilia; cistitis; obstrucción ureteral; ureterocele; cistoscopía.

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ABSTRACT

Eosinophilic cystitis is a rare entity characterized by inflammations with eosinophilic infiltration of the bladder wall. It appears infrequently in children, and there are few case reports in medical literature. This paper presents the case of a 24-month-old patient diagnosed with this condition, the treatment that has been established for it, and a review of the literature.

Keywords: eosinophilia; cystitis; ureteral obstruction; ureterocele; cystoscopy.

RESUMO

A cistites eosinofílica é uma entidade invulgar que se caracteriza pela inflamação com infiltrado eosinofílico da parede vesical. Não é muito frequente na idade pediátrica com mínimos reportes de casos na literatura. Apresentamos um caso de um paciente de 24 meses, a quem se lhe diagnostica dita patologia, o manejo instaurado e uma revisão da literatura. Métodos diagnósticos moleculares em tuberculoses

Palavras chave: eosinofilia; cistite; obstrução ureteral; ureterocele; cistoscopia.

CASE REPORT

A 24-month-old patient was reported to have symptoms of right hydronephrosis associated with a bladder mass compatible with right ureterocele. Unroofing of the ureterocele was carried out through a cystoscopy; there were no complications. A giant ureterocele was found which protruded and obstructed the bladder neck. Afterwards the patient was reported to suffer from frequent urination, dysuria and urinary retention. An ultrasound was then conducted which reported the presence of bilateral hydroureteronephrosis, marked bladder thickening, and bladder hematoma. Surgery was conducted on the patient and no hematoma was found. Nevertheless, severe inflammation associated with polypoid lesions was found in the bladder neck and floor as well as in both meatuses, which were obstructed (Figures 1 and 2). A frozen section biopsy was performed which informed of a possible bladder sarcoma. Therefore, definitive results from pathological anatomy tests

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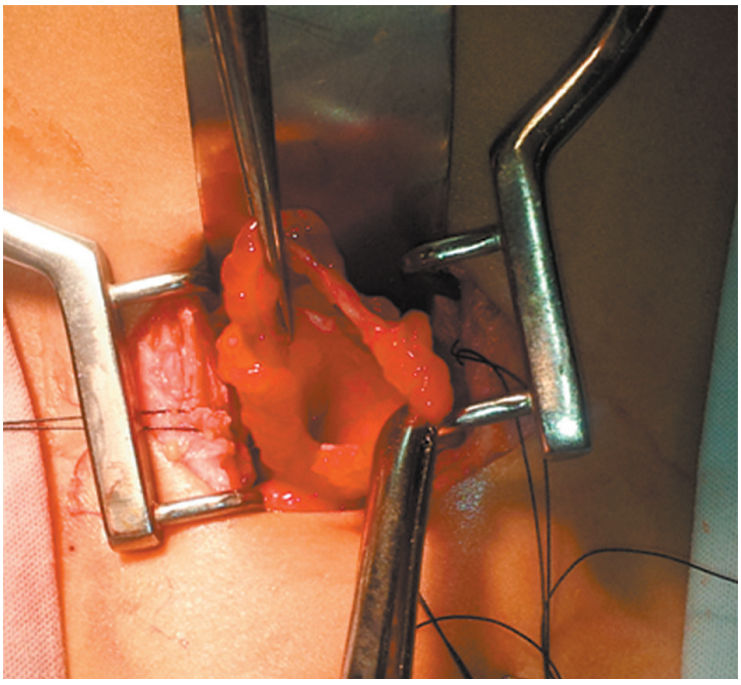

Figure 1. During surgery, a roofless ureterocele (arrow) was observed. However, it was covered by multiple inflammatory polypoid lesions.

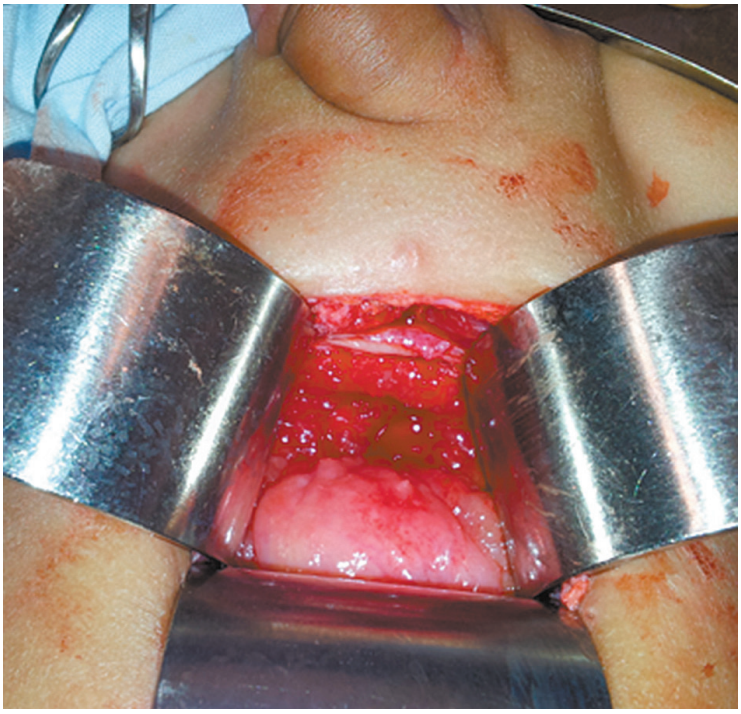

Figure 2. Notice the inflammatory changes in the bladder neck and floor (arrows).

were required in order to define the surgical procedure to be conducted. Likewise, an indwelling urinary catheter was implanted.

The pathology ward finally reported eosinophilic cystitis (Figure 3). Treatment began with high doses of steroids and antihistamines. A cystostomy was performed with a siliconized catheter because of the bladder neck obstruction in order to remove the urethral catheter. The patient showed adequate evolution, thus the cystostomy catheter was closed and urination through the penis was made possible. A control cystoscopy was performed which showed resolution of the inflammatory lesions (Figure 4). After this, the cystostomy catheter was withdrawn and the patient exhibited penis urination without alterations.

LITERATURE REVIEW

Since first being described by Edwin Brown in 1960, eosinophilic cystitis remains a poorly understood clinical entity[1]. Eosinophilic cystitis is a rare form of bladder inflammation characterized by a massive eosinophilic infiltration of the bladder wall. The most common signs and symptoms are pollakiuria, urgent urination, gross hematuria, and hypogastric pain. Compromised ureters can cause hydronephrosis and renal failure. Carcinoma in situ and intractable lower urinary tract obstructions have also been described[2]. Some patients may have a mass effect in their bladders[3]. Male individuals are slightly more affected in comparison to female individuals: 1.3:1. The age of most cases ranges from 18 to 87 years old. Studies conducted on adults have found predisposing factors such as intravesical chemotherapy, transitional cell carcinoma, respiratory disease, bladder obstruction, some medications such as sulfa drugs, cyclophosphamide and warfarin, autoimmune disorders, enteritis, etc[2]. Goble et al. found urothelial eosinophilic infiltration as a response to bladder catheterization[4]. Eosinophiluria and eosinophilia are present in 35-50% of cases. However, eosinophils can be quickly degraded from the urinary sediment, thus it is possible that their presence in the urinalyses is not as constant. Eosinophilic cystitis can progress to fibrosis with bladder retraction; thus it becomes necessary to differentiate it from other conditions such as tuberculous cystitis, interstitial cystitis, and cancer.

Imaging techniques are not diagnostically definitive. There is a wide range of cystoscopic abnormalities, and they can occur with varying degrees of severity ranging from erythema and edema of the mucosa to polypoid, elevated, and fungoid lesions that may even resemble invasive masses[5]. Accurate diagnosis is only possible through biopsies, since they make it possible to see

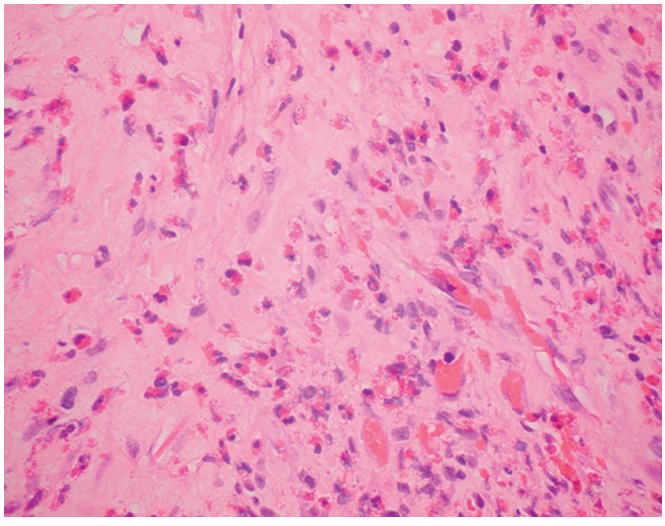

Figure 3. Fibromuscular bladder stroma with numerous eosinophils (arrows) forming true microabscesses. (H & E 10X)

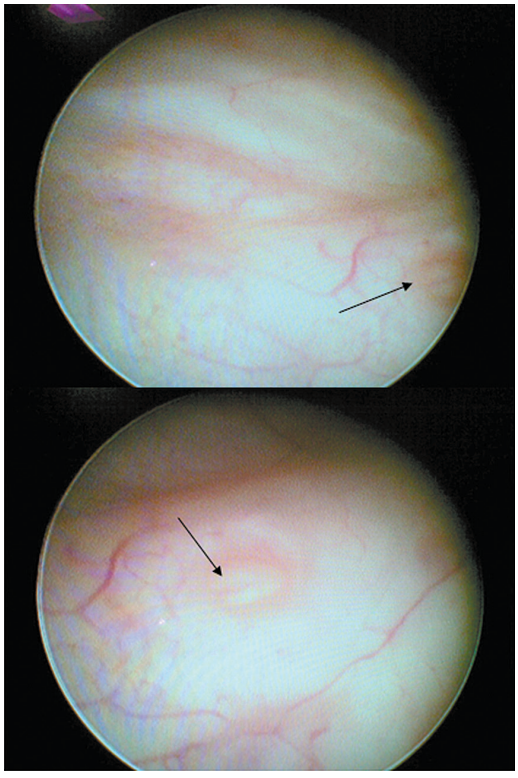

Figure 4. Control cystoscopy. An improvement of the inflammatory lesions was observed. The meatuses became healthy (arrows).

eosinophilic infiltrations in their acute stages, while fibrosis and poor cellularity are predominant in the chronic stages.

This is a rare condition among children, with few cases reported in the literature, where approximately 28 patients below 18 years old are described. In most cases the condition appears as an intravesical tumor with symptoms that are resolved spontaneously or with steroid therapy[6].

The clinical presentation varies[3], whilst the therapeutic approach is anecdotal and functional outcomes range from symptom resolution to intractable bladder dysfunction. Eosinophilic cystitis is the final outcome of allergic cystitis. Its etiology is caused by an IgE-mediated alteration with eosinophil chemotaxis and degranulation of mast cells, however the precise etiology is unknown[7].

Its treatment is empirical[8] and includes the removal of the antigenic stimulus, if known. Anecdotal therapy focuses on inhibiting mast cell degranulation with antihistamines and stabilizing lysosomal membranes with steroids. It is believed that all patients should receive therapy with NSAIDs[9]. Likewise, for refractory cases, either recurrent or with masses, it is useful to use systemic steroids[10]. For cases refractory to the previous therapy, cyclosporin A showed improvement and regression in a pediatric patient[11]. Ureteral obstruction may improve with systemic steroid therapy[12]. For the majority of these patients the disease evolution is benign[13]. There is little data regarding the monitoring of patients with this condition. Studies conducted on adults found that some patients may experience an aggressive and progressive form of the disease that does not respond to medical therapy or transurethral resections, thus requiring nephroureterectomies, partial and total cystectomies, urinary diversions, and neobladder construction[2].

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