

Inflammatory Pseudotumor of the Urinary Bladder in a Child

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Abstract

The inflammatory pseudotumor of the urinary bladder is rare, especially in children. It is a benign proliferative lesion of the submucosal stroma easily mistaken for a sarcoma clinically, so it should be differentiated from a malignant neoplasm. We report the case of bladder inflammatory pseudotumor in a 7-year-old girl.

Key Words: Inflammatory pseudotumor, bladder, child

INTRODUCTION

The inflammatory pseudotumor of the bladder is a benign proliferative lesion of the submucosal stroma easily mistaken for a sarcoma clinically, so it should be differentiated from a malignant neoplasm. Since the first report of inflammatory pseudotumor of the bladder in 1985, a few cases in adult patients have been reported in this Korea.^{1,2} However, only 10 cases involving children have been reported worldwide.³ We report the case of bladder inflammatory pseudotumor in a 7-year-old girl, which presented with a stalk, which is an unusual finding in a child with pseudotumor.

CASE REPORT

A 7-year-old girl presented with frequency, dysuria, and pyuria for 2 weeks unresponsive to antibiotic treatment. Right flank pain and fever accompanied by hematuria were also present for 4 days. Past medical history and family history were both unremarkable. On physical examination, 38.5°C fever was noted, but blood pressure and pulse were within a normal range.

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Lower abdominal tenderness and mild right costo-vertebral angle tenderness were also noted. Urinalysis findings were, pH 5.0, protein (3+), RBC many/HPF, WBC many/HPF. On urine culture, 5×10^3 /ml of *Enterococcus cloacae* were grown. CBC findings were WBC 5,480, Hb 13.3 g/dl, Hct 28.8%, and platelet count of 174,000/ml. Liver and renal functions were both normal. Chest radiographic and EKG



Fig. 1. Findings of urography. The right ureter was mildly dilated and there was egg-sized filling defect in the bladder.



Fig. 2. Gross findings of specimen. The mass was 4×3 cm sized and covered with gray-reddish glistening mucosa. The mass has no capsule, and was partially encapsulated with the transitional epithelium. Most epithelium was detached, and there was hemorrhage and proliferation of capillaries between the inflammatory fibroblasts in the submucosal layer.

findings were negative.

In abdominal ultrasound examination ellipsoid mass lesion with inhomogeneous echogenicity was found. On intravenous pyelography, a hen egg-sized filling defect on the bladder base was observed accompanied by mild to moderate dilatation of the right ureter (Fig. 1). On voiding cystourethrogram, vesico-ureteral reflux grade II on the right ureter and grade I on the left ureter was noted.

We tried cystoscopy under general anesthesia to rule out bladder tumor, ureterocele, blood clot and so forth. On cystoscopy, the bladder was moderately trabeculated with a hen egg-sized well-vascularized smooth-surfaced ellipsoid mass with stalk at the center of trigone. The stalk was 1 cm in length, 1.5 cm in diameter, well-vascularized and connected to the posterior wall of the bladder above the trigone.

Transurethral resection was performed, but bleeding from the tumor mass was severe, and conversion to open cystotomy and tumor excision was required. Mass was 4×3 cm covered with grey-reddish mucosa. Capsule was absent and most epithelium was denuded except for a partial transitional cell covered layer. Spindle-shaped cells accompanied by hemorrhage and inflammatory reaction was distributed between capillary proliferation in the submucosal layer (Fig. 2). Dysplasia of spindle cell was not seen. Mitosis was only rarely seen and atypical mitosis was absent (Fig. 3). Actin, S-100, protein and desmin stains were all

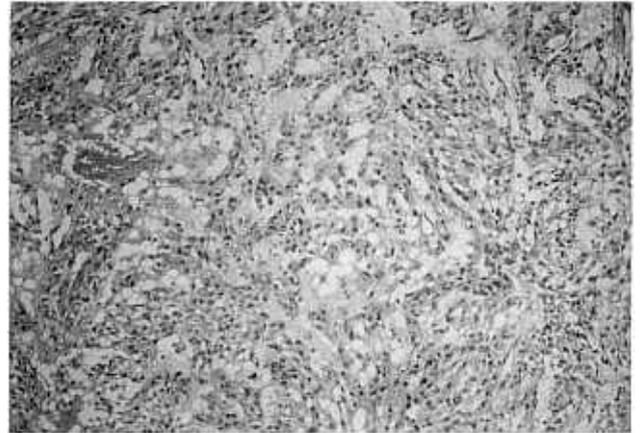


Fig. 3. Microscopic pathologic findings. There was no dysplasia of the spindle cells, and a few mitoses, but no atypical mitoses (×100).

negative.

Voiding symptoms, hydronephrosis and vesicoureteral reflux disappeared after tumor excision. There was no evidence of tumor recurrence at 1 year follow-up.

DISCUSSION

Since the first report of inflammatory pseudotumor of the bladder in 1985, a few cases in adult patients have been reported in this Korea.^{1,2} However, only 10 cases involving children have been reported worldwide.³ It is sometimes called pseudosarcomatous or atypical fibromyxoid tumor or pseudosarcomatous myofibroblastic proliferation,⁴ and most of them initially were misdiagnosed as sarcomas and operated on. The tumors have been found in lung, bronchus, small intestine, mesentery, kidney, spleen, stomach, meninges, spine, thyroid gland, and bladder.⁵ The exact origin is still unclear but there are spontaneously occurring primary cases and operation-related secondary cases. Most occurrences have been found in adults.

Clinically, systemic symptoms of high fever, anemia, hyperglobinemia, polycythemia and urological symptoms of dysuria, frequency, gross hematuria, suprapubic pain are present in inflammatory pseudotumor of the bladder. The growth of tumor is relatively rapid and frequently found as a solitary lesion 1–4 cm in size. It is benign and myxoid stroma fibrinoid cell proliferation are often mistaken

for malignant myosarcoma.^{2,4}

Histologically, invasive growth, abundant cell cytoplasm, multiforme cell division and vessel distribution are characteristic. Spindle-shaped cells in loose myxoid background are also typical. Macrophages, lymphocytes, neutrophils, inflammatory cells are distributed sparsely and foamy histiocytes are aggregated around the mass. Long strap cells are similar to rhabdomyoblasts and can be mistaken for sarcoma.⁶⁻⁸ Electron microscopy, actin, S-100 protein, desmin stains are used to differentiate it from a malignant lesion.

There is no agreed best treatment method due to rarity of the disease. Partial or total cystectomy has been performed. No recurrence has been reported after mass excision or transurethral resection.

The urological symptoms of dysuria, frequency, and gross hematuria have been reported in inflammatory pseudotumor of the bladder. Grossly, the tumor appearance can vary from a pedunculated mass lesion to just thickening of the bladder wall. In this case, irritative bladder symptoms were prominent, and the mass presented with a stalk, which is an unusual finding in a child with pseudotumor.

REFERENCES

1. Lee JH, Kim SJ, Lee YT, Lee JM. A case of inflammatory pseudotumor of the urinary bladder. *Korean J Urol* 1989; 30:944-8.
2. Heo JM, Yoon JB. Inflammatory pseudotumor of the urinary bladder- A case report. *Korean J Urol* 1996;37: 488-91.
3. Enrique F, Ron B, Isaac Y, Gadi H, David C, Marc M, et al. Inflammatory pseudotumor in childhood- a diagnostic and therapeutic dilemma. *Arch Surg* 1991;126: 653-5.
4. Janez L, Andreja Z, Bojan T, Janez J. Sclerosing inflammatory pseudotumor of the urinary bladder in a child. *Am J Surg Pathol* 1992;16:1233-8.
5. George LS, Richard F, Bruce AL, Carleen TB, William B, Thomas AB. Inflammatory pseudotumor (pseudosarcoma) of the bladder. *J Urol* 1989;141:610-2.
6. Lucien EN, Jan MO. Inflammatory pseudotumor of the urinary bladder-possible relationship to nodular fasciitis. *Am J Surg Pathol* 1985;9:366-73.
7. Ro JY, Ayala AG, Ordonez NG, Swanson DA, Babaian RJ. Pseudosarcomatous fibromyxoid tumor of the urinary bladder. *Am J Clin Pathol* 1986;86:583-90.
8. N'Dow J, Brown PAJ, McClinton S, Moffat LEF, Grieve JHK. Case reports-inflammatory pseudotumour of the urinary bladder. *Br J Urol* 1993;72:379-87.