Case Report

Association of Lymphangiectasia with Cystitis Cystica et glandularis mimicking Bladder Tumor: Case Report

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Abstract

Cystitis cystica is a benign lesion and commonly seen with cystitis glandularis and Von Brunn’s nests hyperplasia. Along with its unknown etiology, irritation and inflammation of the bladder mucosa has been held responsible. Even if it is a benign case, the localization of the lesion is of importance as it can involve complications like obstruction. It should be considered in the differential diagnosis of intravesical masses. Medical or surgical treatment alternatives should be evaluated after making a histopathologic diagnosis. In this study, we reported the atypic histopathological diagnosis of cystitis cystica et glandularis presenting with lymphangiectasia by reviewing previous information on the matter.

INTRODUCTION

Cystitis cystica is a proliferative benign lesion forming cystic structures by expanding into lamina propria of Von Brunn’s nests [1]. While it is very rare in men and children, women make up almost all of the cases. In cystitis cystica, nodular changes occur in the bladder mucosa, and there are pearly, pink, brown or yellowish cystic structures protruding into the lumen [2]. To the best of our knowledge, there has been no report of a cystitis cystica et glandularis case presenting with lymphangiectasia in the literature.

CASE PRESENTATION

A 35-year-old male patient applied to our clinic with frequent urination and dysuria ongoing for approximately one year. Upon learning that the patient did not have any additional diseases and had not received any treatment, routine biochemistry, hemogram, urinalysis, direct urinary system graphy, and urinary ultrasonography were performed. Blood, urine, and radiograph tests of the patient showed no pathology, whereas the urinary ultrasonography of the patient revealed a 22x14 mm mass lesion protruding into the lumen in the left lateral wall bladder (Figure 1). In the contrast-enhanced abdominal computed tomography, an mm-calcification inclusive, vegetating solid lesion with lobulated contour was identified in the left posterolateral wall bladder in an approximately 2-cm segment projected from the wall towards the lumen. Cystourethroscopy was planned for the patient considering the probability of malignancy. After completing preoperative preparations, cystourethroscopy was performed under general anesthesia. Transurethral resection was carried out for the bunch of grapes-like lesion monitored lateral to the left ureter orifice in the bladder located in a narrow bed, but spread to a 3-cm region, and showing protrusion to the lumen (Figure 2). The result of the histopathologic analysis was reported as cystitis cystica et glandularis is presenting with lymphangiectasia by reviewing previous information on the matter.
dilated lymphatic canals (lymphangiectasia) (Figures 3, 4, 5). The patient, for whom no treatment besides antibiotherapy was given, is followed closely by our clinic.

**DISCUSSION**

Although it is thought that cystitis cystica is a local immune response developing as a result of inflammation and chronic irritation of the bladder mucosa, its etiology is not fully known [2]. It has been associated with many other factors such as recurrent urinary tract infection, chronic bladder outlet obstruction, neurogenic bladder, and recurrent bladder tumor [3, 4]. Genitourinary system tuberculosis and hormonal changes in the body may be the reasons of this proliferative cystitis. Other factors held responsible except from these are bladder stone, chronic catheterization in paraplegic patients, schistosomiasis, chronic bacterial cystitis, and metaplastic cystitis [5].

In a study evaluating 127 pediatric patients with cystitis cystica, a good correlation has been set between urinary tract infection and cystitis cystica. In addition, it has been stated to present with vesicoureteral reflux and various urinary tract anomalies. Still in the same study, it has been indicated that favorable outcomes have been reached by long-term antibiotic treatment in patients with cystitis cystica. Therefore, cystitis cystica should be considered in the differential diagnosis of patients complaining about recurrent urinary tract infections without any explainable causes [2].

Cystitis cystica is usually seen in the trigonal region, bladder neck, ureterovesical junction, and is rarely seen in the ureters and renal pelvis [2]. Despite being benign histopathologically, they are of importance with respect to provoking urinary system obstructions and related complications in accordance with the site of the lesion [6].

Patients with cystitis cystica are generally asymptomatic and diagnosed incidentally [7]. An image resembling bladder tumor can be seen in ultrasonography. Symptomatic cases can present with frequent urination, hematuria, incontinence, urgency, secondary enuresis, and smelly urine [2, 8]. Patients with lesions in the ureter orifices can present with symptoms such as poor stream, intermittent urination, difficulty to urinate, and residue urinary sensation [9]. Cystitis cystica is a rare but potentially paramount cause of obstruction [3]. Cystitis cystica should be taken into consideration in the differential diagnosis of intravesical soft tissue masses. It has not been clarified whether cystitis cystica is a premalignant lesion or not. As the diagnosis of cystitis cystica is made histologically, biopsies should be taken from suspected regions with cystoscopy. When there is no large lesion in the bladder, its treatment is medical and based upon resolving the underlying irritative factors. In cases of recurrent hematuria or obstruction due to a large mass, surgical treatment might be necessary. The most commonly performed method is
transurethral resection [9]. In recurrent or highly suspicious cases, it should be followed by cystoscopy and urinary cytology [3].

More commonly seen in the adult, black male population, pelvic lipomatosis, whose etiology is not fully known, is characterised by excessive increase of fatty tissue in the perirectal and perivesical regions [10]. Cystitis glandularis is observed in 75% of the cases with pelvic lipomatosis, and their association is also not clear [11]. An image giving rise to the thought of pelvic lipomatosis was not observed in the ultrasonographic and computed tomography (CT) scans of our case.

Cystitis cystica, usually seen with cystitis glandularis and Von Brunn’s nests hyperplasia [12]. Cystitis cystica, partly presenting with cystitis glandularis, was determined in the wake of the histopathological evaluation of our case. Although many publications regarding intestinal lymphangiectasia were found during literature review, a case of cystitis cystica and glandular is presenting with lymphangiectasia in the bladder was not come upon. Therefore, there is not sufficient data in order to explain the association of cystitis cystica and glandularis with lymphangiectasia in the bladder. However, we are of the opinion that lymphangiectasia may develop as a result of inflammatory processes, lymphatic obstructions, and stasis. No association was reported between either cystitis cystica et glandularis or intestinal metaplasia and pelvic lipomatosis. Although intestinal metaplasia can be seen in these cases, it was not present in our patient.

The aspects that make our case different from the more common cases are that the gender of our patient was male, which is a gender encountered less often in this disease, dilated lymphatic canals (lymphangiectasia) accompanying cystitis cystica et glandularis in the histopathologic evaluation, and its resemblance of a bunch of grapes macroscopically. The patient is still followed closely, and pathology was not detected in the bladder during the cystoscopy performed in the second year.

REFERENCES