



Case report

Severe obstructive symptoms and urinary bladder mass due to cystitis glandularis: A very rare case report in children

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ABSTRACT

Cystitis Glandularis (CG) is an unusual proliferative disease of the bladder. This condition was associated with chronic inflammation or chronic obstruction. This condition usually presents as microscopic finding and the presence of large macroscopic lesion is a rare feature. Until now, the course of disease from transitional to cystitis glandularis is still unclear and the uncertainty of CG to potentially develop into adenocarcinoma has once been documented. Herewith, we report our experience with 2 years old boy with cystitis glandularis presenting with LUTS obstructive symptoms, hematuria and bladder mass. Ultrasound examination revealed bilateral hydronephrosis with hydroureter and bladder wall thickness suggesting the sign of obstruction and chronic inflammation. Cystoscopy examination was performed to ensure the diagnosis with the result revealing protruding mass partially obstructing the bladder trigone, both ureteral orifice and posterior urethra. Transurethral resection was performed and the administration of COX-2 inhibitor and oral steroid therapy were given. Post-operative course was uneventful with the improvement in symptom and uroflowmetry revealed promising result. This case represented an entity of rare and interesting case of cystitis glandularis causing severe obstructive symptoms and urinary bladder mass which appropriate therapy of endoscopic intervention, COX-2 inhibitor and oral steroid resulted in promising outcome. Follow up of 1 year resulted in reduced LUTS symptoms such as straining and difficulty of urination.

1. Introduction

Cystitis glandularis (CG) was first described by Morgagni et al. in 1761. Cystitis glandularis is an unusual proliferative disease of the bladder. This case represented metaplastic lesion in the bladder which is confirmed from histopathological appearance [1]. CG was most commonly found in pediatric population and rarely occurred in adult. This condition was associated with chronic inflammation or chronic obstruction [2–4]. Chronic inflammation will result in hyperproliferation of the bladder wall. The most common symptoms presented in patient with cystitis glandularis are hematuria, irritative or obstructive voiding symptoms [1]. CG is usually a microscopic finding which large visible macroscopic lesion is a rare manifestation. However, several reported cases have demonstrated the case of CG manifested as bladder mass [5]. Histologically, cystitis glandularis would be seen as a cystic sac consisted of columnar or cuboidal cells as a result from proliferation in Von Brunn Nest's. This disease possesses the characteristic of transitional cell that undergoes glandular metaplasia. Until now, the course of disease from transitional cell to cystitis glandularis is still

unclear. The patient with cystitis glandularis was recommended to undergo routine endoscopic evaluation. This is due to several reported cases mentioning that cystitis glandularis will transform into adenocarcinoma. Even in a study done by Immergue and Cottler showed that cystitis glandularis play role in the occurrence of bladder adenocarcinoma [2,3]. Moreover, the need to conduct early and aggressive treatment is controversial. Several previous studies suggested the use of transurethral resection and cystoscopy to evaluate adenocarcinoma changes of the bladder [6]. This work has been conducted in accordance with SCARE and PROCESS guideline [7,8]. Herewith, we report our experience in the management of 2 years old boy with cystitis glandularis presenting with LUTS obstructive symptoms, hematuria and bladder mass.

2. Presentation of case

A 2 years-old boy presented in the urology OPD with the symptom of difficulty in urination. This symptom was first felt since the patient was 3 month years old. The patient complained pain during urination.

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However, the parent was late to realize that their child complained pain everytime the patient urinates. Moreover, the recurrent hematuria was present. The history of stone expulsion, and previous LUTS symptoms were denied. The patient had normal antenatal history, and routine antenatal visit. Patient had normal history of immunization. There was no family history with the same symptoms or family history of malignancy. Patient had previous history of scrotal hernia and rectal prolapse. The patient underwent the procedure of hernia repair. Laboratory and urine examination were unremarkable. The upper and lower abdominopelvic ultrasound as shown in Fig. 1 revealed abnormality in the bladder with detrusor muscle hypertrophy suggesting the presence of chronic cystitis. This irregular bladder wall thickening represented in the size of 2 cm. Solid mass infiltrating posterolateral wall of the bladder with positive CDUS (Colour Doppler Ultrasound) was also identified. Moreover, severe bilateral hydronephrosis with hydroureter were also observed. Intravenous Pyelography examination revealed the presence of cystitis with good kidney excretion function without signs of obstruction and stone in the ureter (Fig. 2). The Voiding Cysto-Urethrogram (VCUG) examination revealed the sign of chronic cystitis without the presence of Vesicoureteral Reflux (VUR) as shown in Fig. 3. Serial cystoscopy was performed with all the results revealing the presence of papilar mass in prostatic urethra, bladder neck, bladder trigone and posterior bladder. The mass was also observed partially obstructing the bladder trigone, both ureteral orifice and posterior urethra. Transurethral biopsy and resection were then performed with the pathological result revealing cystitis glandularis. All three serial cystoscopies were detailly showed in Fig. 4. Urodynamic evaluation revealed the result of low bladder capacity, overactive bladder, and cystitis as shown in Fig. 5. Uroflowmetry test showed the result of voiding time 24 s, with voided volume 18 ml, Qmax 3.2 ml/s and PVR of 38 ml. Moreover, the test of cyclooxygenase-2 (COX-2) antibody was performed with positive result. The patient then underwent oral COX-2

therapy with celecoxib 50 mg. Due to non-improvement of symptom, and after multidisciplinary consultation, oral prednisolone with the dose of 1×2.5 mg was given. Follow up of 1 year resulted in reduced LUTS symptoms such as straining and difficulty of urination. Uroflowmetry evaluation after therapy also revealed improvement with Qmax of 10.4 ml/s, voided volume of 39 ml, and PVR of 54.1 ml. The comparison of uroflowmetry result before and after therapy was shown in Table 1.

3. Discussion

Cystitis Glandularis (CG) is a rare but benign disease causing hyperproliferation of the mucus-producing glands from the mucosa and submucosa of bladder epithelium. Normally, the pathological finding was microscopic. However, macroscopic lesion may also be found although it is very rare. Because the potential of malignancy is uncertain, the management of cystitis glandularis has not been established and is still debatable. The management of CG ranged from transurethral resection to a more extensive surgical management especially in the case of severe or high recurrency [1]. Previous reported case had provided the evidence of malignancy from CG. Since progression of CG into adenocarcinoma has been documented, serial cystoscopy along with bladder biopsy was also recommended [7]. However, another study also revealed that there is no evidence in which cystitis glandularis will increase the risk of malignancy. Another study from Corica et al. also reported that intestinal metaplasia is not a risk factor from bladder malignancy [8]. Herewith, we report a rare case of 2 years old boy with symptomatic cystitis glandularis causing severe obstructive urinary symptoms and bladder mass. Eventhough there are already more than a hundred of reported cases of cystitis glandularis, a CG case causing severe obstructive symptoms and bladder mass is still a very rare entity in previous literature.

Microscopically, there are two types of cystitis glandularis which are

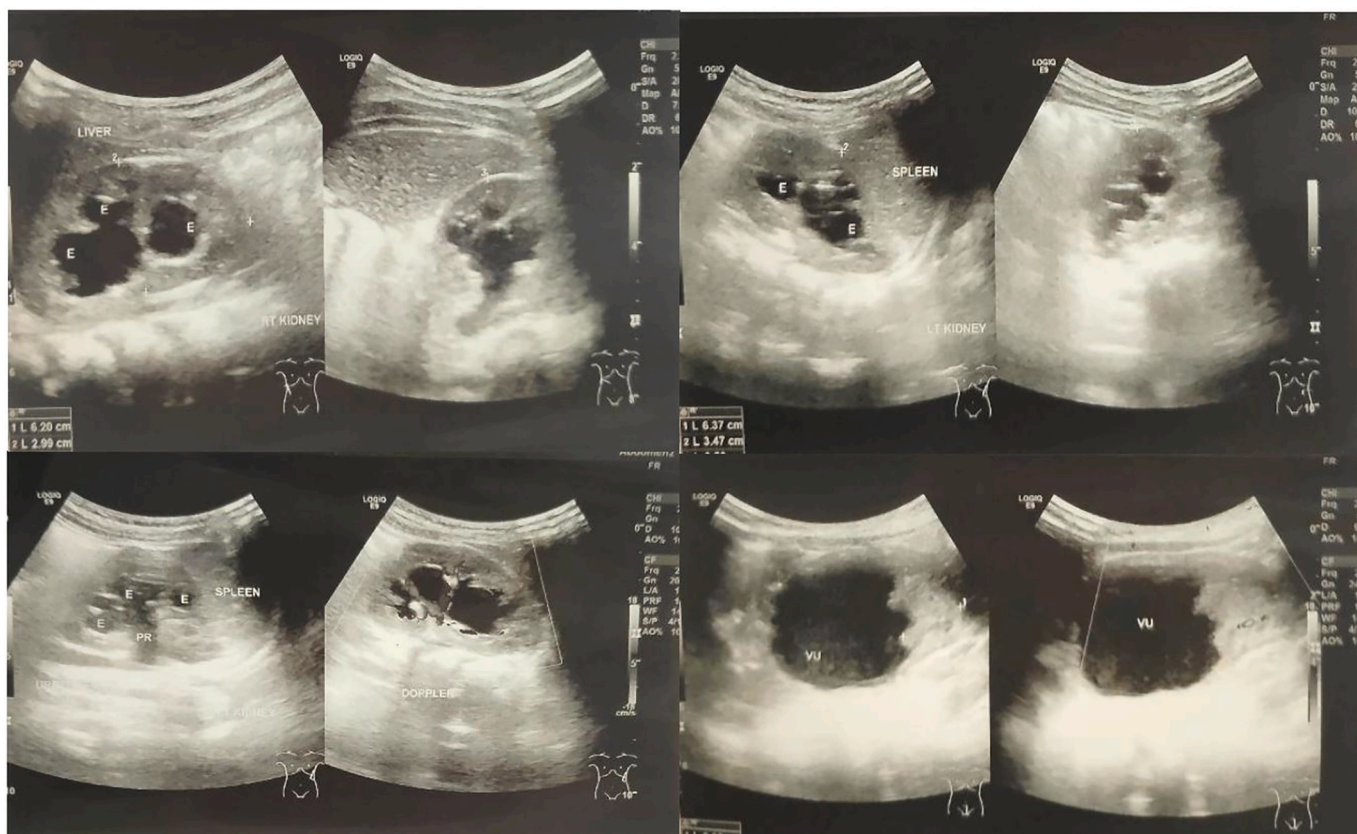


Fig. 1. Ultrasound examination showed the sign of obstruction and inflammation.

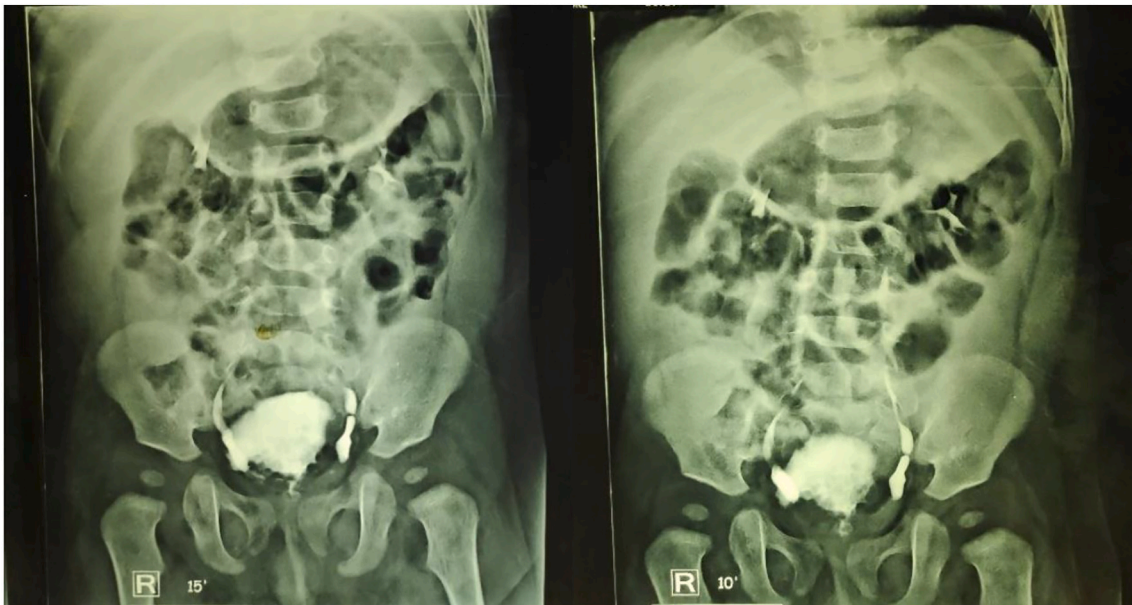


Fig. 2. Intravenous Pyelography examination revealed the presence of cystitis with good kidney excretion function without signs of obstruction and stone in the ureter.



Fig. 3. Voiding Cystourethrogram revealed the result of chronic cystitis without VUR.

common type and intestinal type. Intestinal type possessed the similarity with adenocarcinoma. Several cases even demonstrating the progression from cystitis glandularis into adenocarcinoma [4]. In this case, histological examination revealed stromal cell lined with columnar cells with the surrounding of inflammatory cells which suggested the common type of cystitis glandularis.

Cystitis glandularis usually do not cause significant symptoms however sign and symptoms which may occur were frequency, urgency, dysuria and hematuria [2,9]. In relation to this case, the patient complained LUTS obstructive symptom including hesitancy, straining and difficulty of urination. The obstructive symptom may occur when the mass was found in the bladder trigone [10]. Other symptom from the patient was recurrent hematuria. The presenting symptom of hematuria was also reported in previous report [11]. The history of recurrent urinary tract infection was also found in this patient. This finding was in accordance with previous literature which mentioned that cystitis glandularis occurs as a result of chronic inflammation in the bladder in which the patient in this case possess a history of recurrent urinary tract infection [12]. Physical examination including both right and left kidney, and suprapubic revealed no sign of abnormalities. However, examination in anorectal region showed the presence of rectal prolapse

which interdepartmental consultation to pediatric surgery was conducted regarding this condition. Rectal prolapsed in this patient was believed due to excessive straining in order to be able to urinate in CG patient.

Cystoscopy examination revealed mass in the bladder trigone and bladder neck. Ultrasonography examination showed the presence of bilateral severe hydronephrosis, bilateral hydroureter, and bladder wall thickness with irregular shape suggesting the occurrence of chronic inflammation. This result was in accordance with history of recurrent urinary tract infection. The presence of hydronephrosis in this patient was due to bladder mass (cystitis glandularis) which was commonly found in bladder trigone and bladder neck causing the patient in difficulty of urination and straining when urinate. The finding of hydronephrosis in this patient is in accordance with the another report which has proven that CG may cause urinary obstruction and even leading to acute renal failure [13,14]. Cystitis glandularis usually presented as microscopic finding and urinary bladder mass is extremely rare in the literature [1,15]. Therefore, the finding of bladder mass mimicking the characteristic of bladder tumor was a feature which makes this case rare and interesting.

The treatment of cystitis glandularis according to the literature is by

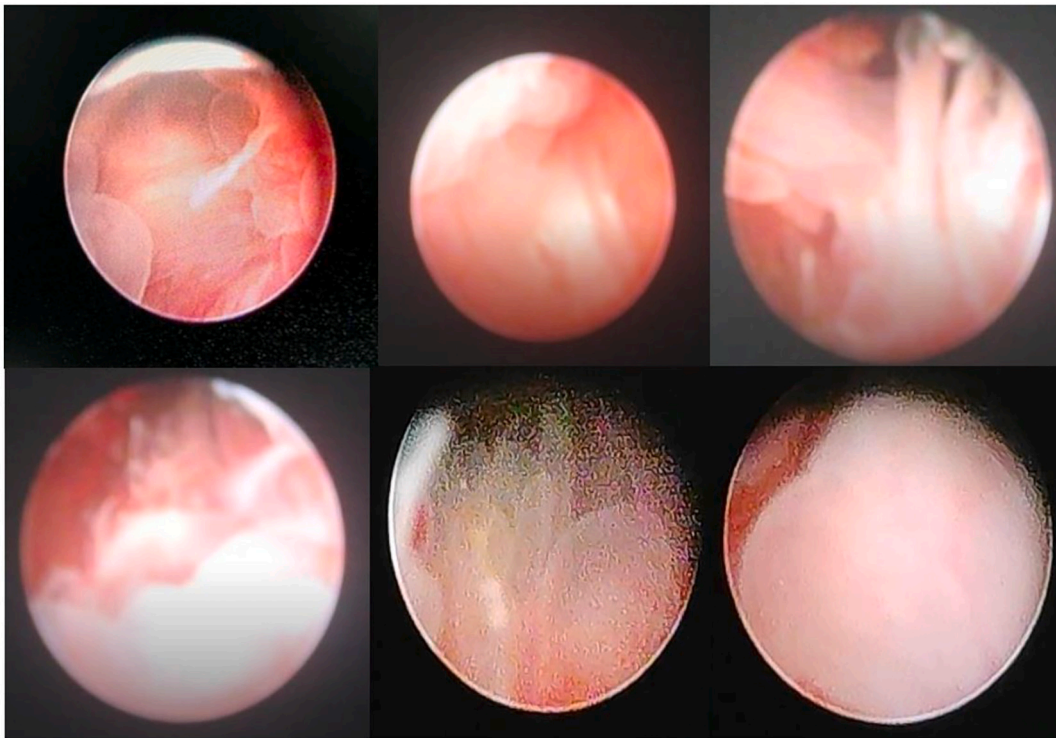


Fig. 4. Serial cystoscopy evaluation revealed mass partially obstructing the bladder trigone, ureteral orifice and posterior urethra.

performing transurethral resection, removing obstruction, reducing the inflammation and the administration of long term antibiotic [2,9]. In this case, patient performed three times of Transurethral Resection of the Bladder (TURB) to resect the bladder mass. Aside from management of inflammation and obstruction as risk factors causing cystitis glandularis including transurethral resection of the bladder lesion, various other treatment modalities have also been reported in the literature. The use of COX-2 inhibitor and oral steroid has been demonstrated [4,12]. In relation to this case, after resection was performed, the patient was then given oral therapy using COX-2 inhibitor of Celecoxib 50 mg. The treatment of COX-2 inhibitor was given after COX-2 antibody examination was tested positive. This therapy was according to the literature which stated that intestinal metaplasia possesses the similarity with colon mucosa in terms of morphology and gene expression. COX-2 inhibitor such as celecoxib was proven to be effective in prevention of colorectal adenoma [12]. However, after the patient received oral treatment of celecoxib for approximately 4 months, evaluation using uroflowmetry and PVR suggested that the obstruction due to cystitis glandularis has not been resolved and the patient did not respond upon the therapy of celecoxib. Therefore, the need for other therapy was mandated. Another option of therapy for cystitis glandularis in this patient are the administration of oral steroid such as prednisolone [4,9,12,16]. The patient then received oral therapy of prednisolone. The therapy of oral prednisolone was in accordance to the current literature which stated that a resistant case of cystitis glandularis to endoscopic intervention could be given with oral steroid therapy [4]. In this case, the patient was given oral prednisolone 10 mg once daily. The improvement of symptom relieving was seen after three months of therapy. The therapy was then continued with reduced dose of 1×5 mg prednisolone in which improvement of symptom and cystoscopy were seen in this patient.

The therapy of oral prednisolone was then continued with the dose of 2.5 mg for 1 year and the improvement of LUTS symptom was observed. The urination of the patient was easier without excessive straining. After the administration of oral prednisolone for 1 year, the patient felt ease to urinate and the frequency of occurrence for rectal prolapse was reduced.

This result was confirmed with the improvement from uroflowmetry and PVR. Table 1 depicted the comparison of uroflowmetry and PVR result before and after treatment.

4. Conclusion

Cystitis glandularis is a disease associated with inflammation or chronic obstruction. The symptoms ranged from asymptomatic to symptomatic such as irritative LUTS, obstructive LUTS or hematuria. This condition usually presents as microscopic finding and the presence of large macroscopic lesion is a rare feature. Medical treatment is considered if this condition does not produce symptoms. However, in the presence of obstructive or irritative voiding symptoms, the treatment of cystitis glandularis may require transurethral resection to relieve the obstruction or inflammation. Other alternative therapy which could be given includes the administration of oral COX-2 inhibitor (celecoxib) or oral steroid administration such as prednisolone specifically in a case of cystitis glandularis resistant to endoscopic intervention.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethical approval

Ethical approval has been acquired in this study.

Funding

No funding was granted in this study.

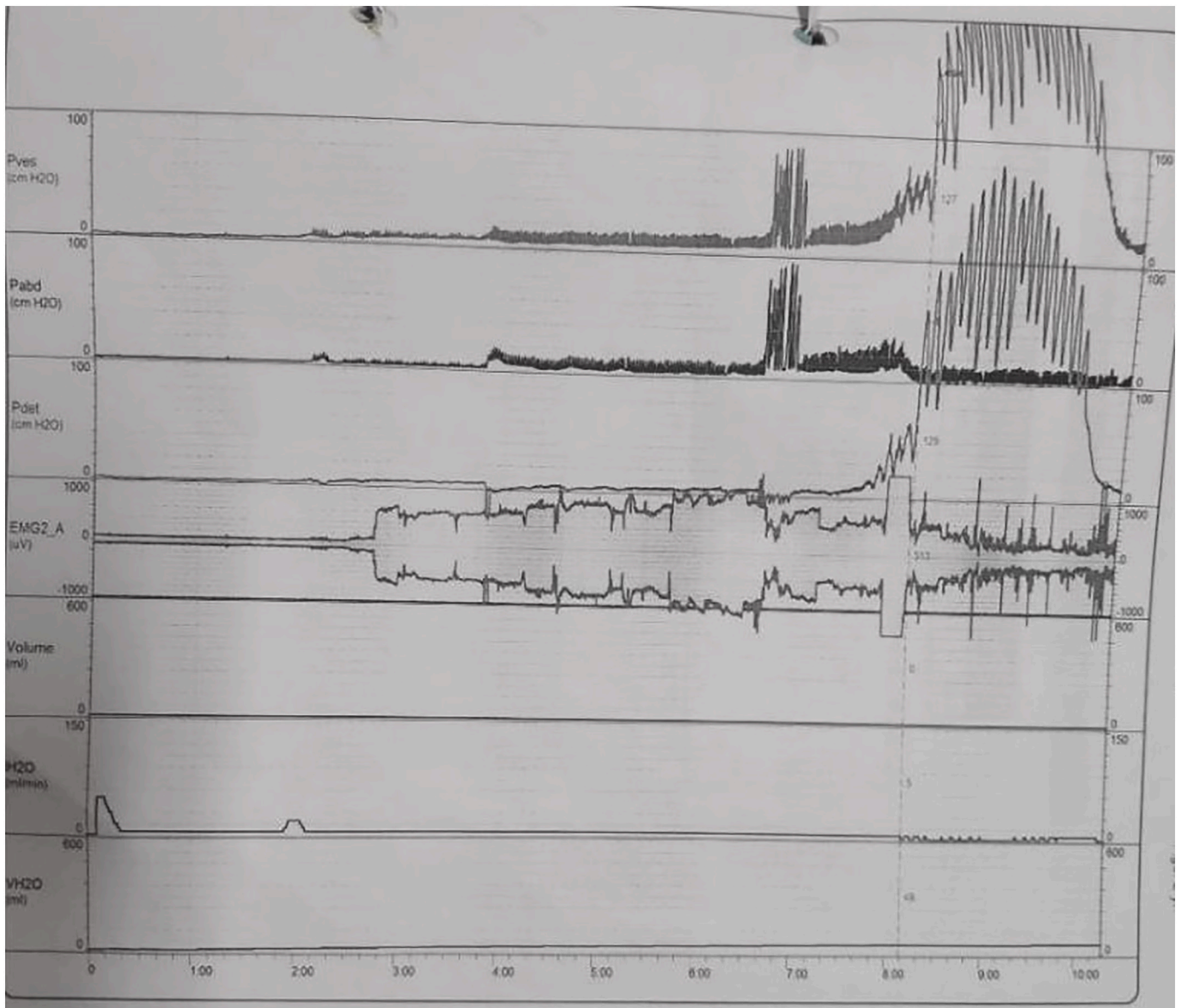


Fig. 5. Urodynamic evaluation revealed low bladder capacity, overactive bladder, and cystitis.

Table 1

The comparison of uroflowmetry before and after treatment.

	Before therapy	After therapy
Voided volume (ml)	18	39
Q max (ml/s)	3,2	10,4
PVR	38	54,1

Author contribution

Conceptualization – AK, STJ; Data curation – AK, IAR; Materials – AK, STJ; Formal Analysis – AK, IAR; Investigation – AK, IAR; Methodology – AK, IAR; Supervision – STJ; Writing original draft – AK, IAR; Writing, review and editing – IAR, STJ.

Guarantor

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Research registration number

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Provenance and peer review

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Declaration of competing interest

No conflict of interest in this study.

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