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ABOUT COVER

Editorial Board Member of *World Journal of Clinical Cases*, Pietro Scicchitano, MD, Professor, Research Scientist, Department of Emergency and Organ Transplantation, School of Medicine, University of Bari, Bari 70124, Italy. piero.sc@hotmail.it

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Pelvic lipomatosis with cystitis glandularis managed with cyclooxygenase-2 inhibitor: A case report

Li-Cai Mo, Song-Zhe Piao, Hai-Hong Zheng, Tao Hong, Qin Feng, Mang Ke

ORCID number: Li-Cai Mo 0000-0002-4272-3210; Song-Zhe Piao 0000-0002-4219-0645; Hai-Hong Zheng 0000-0003-2914-0413; Tao Hong 0000-0001-8574-4911; Qin Feng 0000-0002-2564-7666; Mang Ke 0000-0002-5547-6351.

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Li-Cai Mo, Song-Zhe Piao, Tao Hong, Qin Feng, Mang Ke, Department of Urology, Taizhou Hospital of Zhejiang Province Affiliated with Wenzhou Medical University, Taizhou 317000, Zhejiang Province, China

Hai-Hong Zheng, Department of Pathology, Taizhou Hospital of Zhejiang Province Affiliated with Wenzhou Medical University, Taizhou 317000, Zhejiang Province, China

Corresponding author: Mang Ke, MD, Doctor, Professor, Department of Urology, Taizhou Hospital of Zhejiang Province Affiliated with Wenzhou Medical University, No. 150 Ximen Street, Taizhou 317000, Zhejiang Province, China. kem@enzemed.com

Abstract

BACKGROUND

Pelvic lipomatosis (PL) is a rare benign condition with characteristic overgrowth of histologically benign fat and invasion and compression of pelvic organs, often leading to non-specific lower urinary tract symptoms (LUTS). Approximately 40% of patients with PL have cystitis glandularis (CG). The cause of PL combined with CG is poorly understood, and there is currently no effective treatment. Refractory CG with upper urinary tract obstruction even requires partial or radical bladder resection.

CASE SUMMARY

In this case, a patient suffering from PL with CG was treated by transurethral resection of bladder tumour (TUR-BT) and oral administration of celecoxib, a selective cyclooxygenase-2 (COX-2) inhibitor. The LUTS were alleviated, and the cystoscopy results improved significantly. Immunohistochemistry showed up-regulated COX-2 expression in the epithelium of TUR-BT samples, suggesting that COX-2 may participate in the pathophysiological process of PL combined with CG.

CONCLUSION

We report for the first time that celecoxib may be an effective treatment strategy for PL combined with refractory CG.

Key Words: Pelvic lipomatosis; Cystitis glandularis; Cyclooxygenase-2; Celecoxib; Lower urinary tract symptoms; Case report

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Core Tip: Pelvic lipomatosis (PL) is a rare benign condition with characteristic overgrowth of histologically benign fat and invasion and compression of pelvic organs. The cause of PL with cystitis glandularis (CG) is poorly understood, and there is no effective treatment. This paper reports a patient suffering from PL with CG who was treated by transurethral resection of bladder tumour and oral administration of celecoxib. The lower urinary tract symptoms were alleviated, and the cystoscopy results improved significantly. We report for the first time that celecoxib may be an effective treatment strategy for PL combined with refractory CG.

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INTRODUCTION

Pelvic lipomatosis (PL) is a rare proliferative disease that was first identified by Fogg *et al*[1] early in 1968. Nevertheless, no more than 150 cases have been reported in the English-language literature[2]. PL is characterized by the excessive and diffuse growth of histologically benign fat in the perirectal and perivesicular spaces. It has been reported that more than 75% of PL patients suffer from proliferative cystitis, and an additional 40% of these patients show cystitis glandularis (CG) as a comorbidity[3]. There is currently no effective treatment for PL combined with CG. Approximately 40% of patients may develop obstructive uropathy within 5 years after diagnosis, probably leading to renal failure[4]. Therefore, surgical intervention is often necessary in patients with upper urinary tract obstruction, including ureteral reimplantation, urine drainage (indwelling catheter), and partial or total bladder resection, among others. However, the efficacy of these strategies is not ideal, and the quality of life of patients is seriously affected by the above surgical interventions. A large number of studies have reported that cyclooxygenase-2 (COX-2) is highly expressed in CG tissues[5,6]. Herein, we report the first case of a patient with PL combined with CG who received the COX-2 inhibitor celecoxib in combination with transurethral resection of bladder tumour (TUR-BT), which improved his condition significantly. Additionally, his imaging characteristics were reassessed.

CASE PRESENTATION

Chief complaints

A 49-year-old man complaining of lower urinary tract symptoms (LUTS) with haematuria for 1 year was taken to Taizhou Hospital of Zhejiang Province Affiliated with Wenzhou Medical University, China. The core lower urinary tract symptom score (CLSS) was 11 points.

History of present illness

The patient reported that he had suffered from LUTS and haematuria for 1 year.

History of past illness

The patient had no history of other illness.

Personal and family history

The patient did not smoke or drink, and had no relevant family history.

Physical examination

No positive manifestation was found on prostate examination.

Laboratory examinations

Urine sediment microscopic examination revealed urinary occult blood (ERY) 1+. The results of routine blood test, urine cultures, and renal function tests were normal. Urine cytology revealed no atypical cells.

Imaging examinations

Ultrasound of the urinary system revealed irregular thickening of the bladder triangle where burr-like protrusions were visible and no hydronephrosis (Figure 1A). Contrast-enhanced computed tomography (CT) of the pelvis showed that the bladder wall was thickened, especially the posterior wall and the bladder triangle. Polypoidal growth inside the bladder was noted (Figure 1B). Moreover, no obvious dilatation was found in the kidneys or ureters (Figure 1C). Cystoscopy and biopsy showed bladder lesions in the trigonum vesicae, cervix vesicae, and both sides of the ureteral orifices (Figure 2A and 2B). Histopathological examination suggested CG.

The patient underwent TUR-BT under general anaesthesia with endotracheal intubation. The patient was followed for 3 mo after the first TUR-BT procedure. At this time, the diagnosis of PL was confirmed by CT. The biopsy, unfortunately, had not been performed without the patient's permission. CT of the urinary tract showed that the bladder morphology had changed from an elliptical shape to a pear shape (Figure 1D and F). Additionally, changes from no dilatation of the kidneys or ureters to bilateral dilatation of the renal pelvis, calyces, and ureters was observed and was more obvious in the left upper urinary tract (Figure 1C and E). An increased fat density around the bladder and rectum was noted, as well as compression of the rectum (Figure 1F). The angle between the bladder and seminal vesicle in the axial image (\angle BOS) changed from 49.5° to 89.5° and the diameter of the rectal axial cross-section changed from 2.08 cm to 1.76 cm (Figure 1D and F).

FINAL DIAGNOSIS

Based on the findings of the CT and histopathological examinations, a provisional diagnosis of PL combined with CG was made.

TREATMENT

The patient underwent TUR-BT under general anaesthesia with endotracheal intubation. Besides from the bladder lesions on both sides of the ureteral orifices, the others were completely removed. The bladder lesions on both sides of the ureteral orifices were left *in situ* during the operation, and JJ stents were placed prophylactically. Pathology showed von Brunn nest hyperplasia and goblet cells secreting considerable mucin, consistent with intestinal epithelial CG (Figure 2C).

At the 1-mo follow-up, the JJ stents on both sides were removed. Left renal colic occurred 3 mo after surgery, with abnormally increased renal function indexes (serum creatinine: 1.63 mg/dL; estimated glomerular filtration rate: 49 mL/min/1.73 m²). Cystoscopy indicated the recurrence of CG. We repeated TUR-BT 6 mo after the initial operation, with a similar pathological result, namely, CG. The patient was followed for 3 mo after the second TUR-BT procedure. The CLSS score was 12. The cystoscopy results suggested limited benefit of the above treatment on CG.

Re-examination of renal function revealed that the serum creatinine level was 1.92 mg/dL and the estimated glomerular filtration rate was 40 mL/min/1.73 m². According to cystoscopy results and CT findings, CG was determined to have recurred, with upper urinary tract obstruction on both sides. Therefore, a third TUR-BT procedure and ureteroscopy on both sides were carried out, only revealing the recurrence of CG in the bladder. However, the stricture of the left ureteral orifice made it difficult to insert the flexible ureteroscope (fUR) into the ureter. After using the guide wire to pass through the twisted section of the left ureter, the fUR was placed. Small stones were found in the lower part of the left ureter where a 12/14-Fr ureteral access sheath (UAS) was placed, and three small stones were removed through the UAS using collection basket. There were no obvious abnormalities of the right ureter. One month after the third operation, the JJ stents on both sides were removed together. Postoperatively, the patient was orally given the COX-2 inhibitor celecoxib for 6 mo (200 mg twice daily; Pfizer, United States). Although the indications for celecoxib do not include CG, it can act as an anti-inflammatory agent and postope-

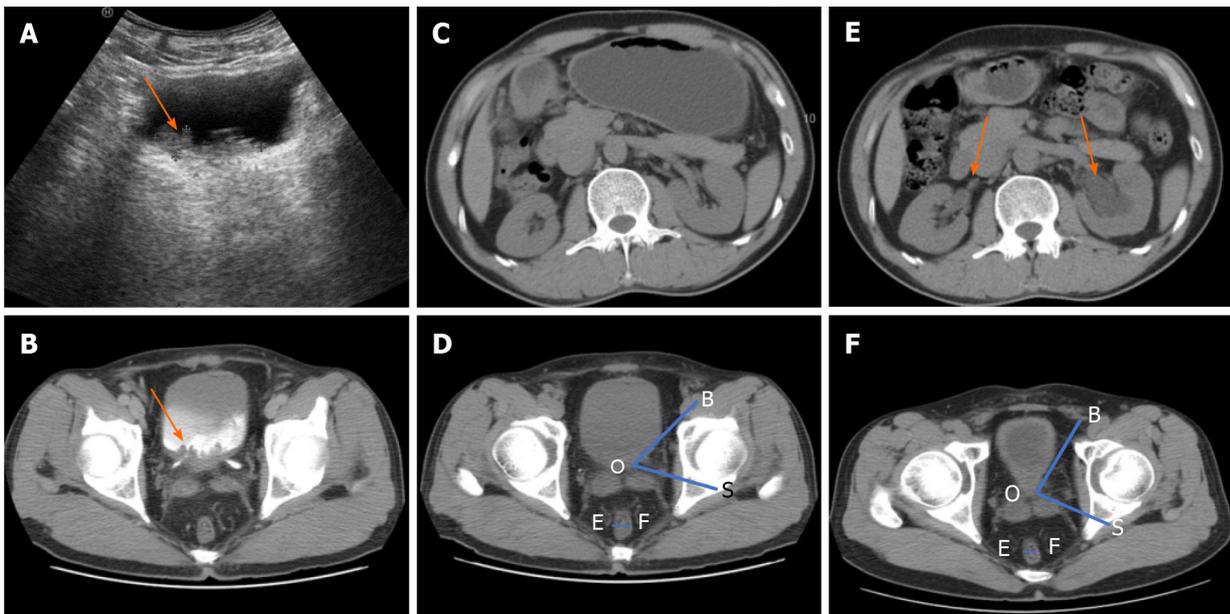


Figure 1 Imaging findings of cystitis glandularis with pelvic lipomatosis. A: Ultrasound examination showed that the mass was located in the triangular region of the bladder, with a burr-like protrusion and irregular thickened wall of the triangular region of the bladder; B: On first admission, computed tomography (CT) of the urinary system suggested thickening of the bladder wall, mainly on the posterior wall and the triangle area, and the mastoid process was observed inside the cavity. Mild to moderate continuous enhancement was noted in the delayed phase; C: Urinary CT scan for the first time revealed no hydronephrosis in the renal pelvis and ureter; D: Angle between the bladder and seminal vesicle (ABS) in the axial cross-section (\angle BOS) and the axial cross-sectional diameter of the rectum (EF) in CT at the first visit. The bladder morphology was elliptical; E: CT was reviewed 3 mo after the second transurethral resection of bladder tumour (TUR-BT). The pelvises, calyces, and ureters on both sides dilated, especially on the left side; F: CT review 3 mo after the second TUR-BT: ABS in the axial cross-section (\angle BOS) became larger and the axial cross-sectional diameter of the rectum (EF) became smaller. The adipose density around the bladder and around the rectum increased, the rectum was compressed, and the bladder shape was inversely pear-shaped.

rative analgesic. The patient was informed about the necessity for and the risk of the off-label use of celecoxib, and his consent was obtained.

OUTCOME AND FOLLOW-UP

The patient was followed at 6 mo and 1 year after the treatment. No CG recurrence was found cystoscopically (Figure 2E and F). The CLSS was decreased significantly at 6 mo and 1 year after treatment (9 and 8 points, respectively) compared to baseline (11 points). At the last follow-up, the patient's clinical manifestations were stable, with no recurrence of CG on cystoscopy. The CLSS was 8 points, haematuria was absent, and the serum creatinine level was stable at 1.63 mg/dL (estimated glomerular filtration rate: 49 mL/min/1.73 m²), although subsequent CT scans revealed PL with obstructive uropathy. We performed a retrospective analysis of the patient's TUR-BT tissue samples, and applied COX-2 antibody to detect the expression level of COX-2 in the CG tissue by immunohistochemistry (Figure 2D). The staining steps were conventional dewaxing and antigen retrieval. Primary antibody (COX-2 monoclonal antibody; Clone No. 3G2B9, Proteintech Group, United States) was added. The next day, we added secondary antibody and incubated the samples for 30 min. Then, we performed DAB staining, hematoxylin counterstaining, and sealing with neutral gum.

DISCUSSION

PL is a rare disease of benign proliferation. Because of the subtle symptoms of early PL, some elderly male patients may be missed, and the incidence may be underestimated[7]. Subsequently, patients with advanced disease are troubled with haematuria, unendurable LUTS, or gastrointestinal symptoms, such as constipation, nausea, lower abdominal pain, or progressively increasing back pain. Severe hydronephrosis and renal insufficiency may have developed in some patients by the time of diagnosis. Approximately 1/3 of patients with PL exhibit hypertension, and some patients exhibit uremia due to renal failure. The patient in the present case

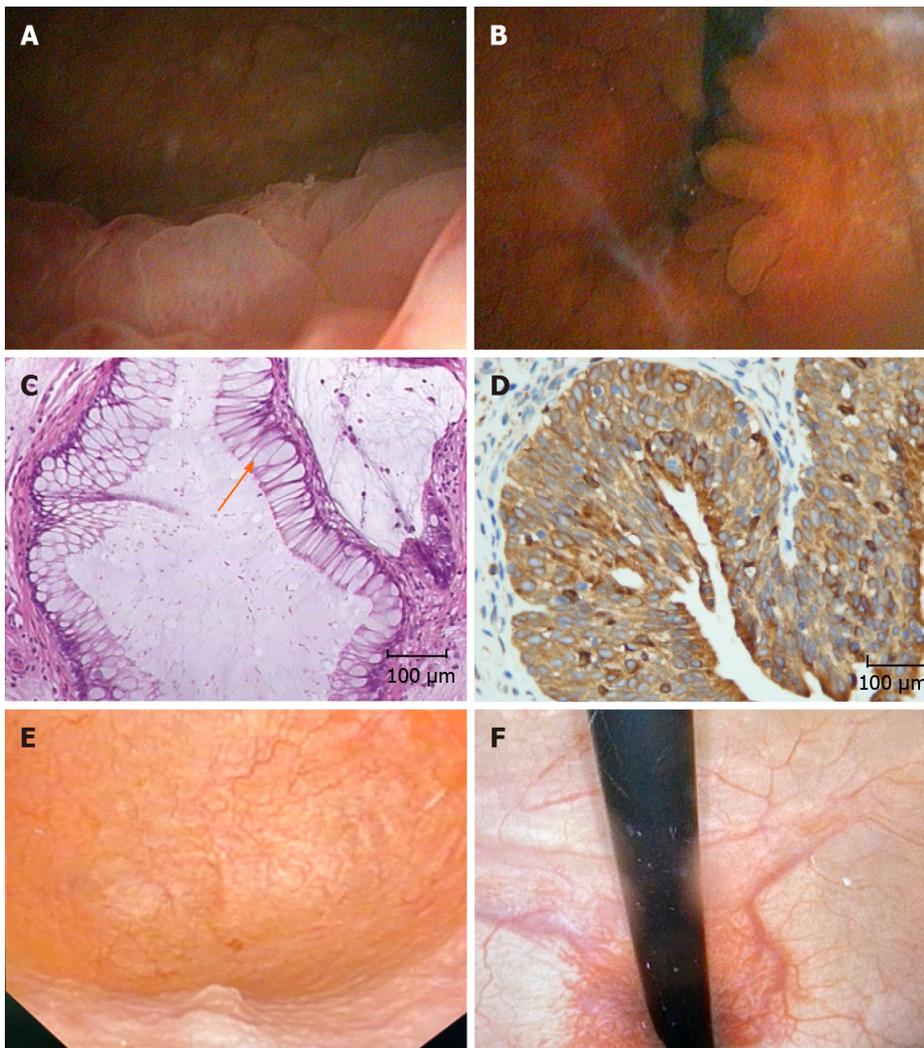


Figure 2 Cystoscopy and pathological results. A and B: Bladder lesions in the trigonum vesicae, cervix vesicae (A), and both sides of ureteral orifices (B) were observed at the first visit; C: The initial histopathological analysis after transurethral resection of bladder tumour showed considerable goblet cells (shown by arrows) in the bladder epithelium; D: Immunohistochemical analysis of cystitis glandularis showed positive expression of cyclooxygenase-2 (COX-2) in the cytoplasm. The pathological results showed that it was intestinal glandular cystitis; E and F: Negative recurrence of bladder tumor was observed cystoscopically after 6-mo application of COX-2 inhibitor.

presented for the first time without significant hypertension but with significant LUTS and gross haematuria. However, there were no abnormalities on CT, indicating that upper urinary tract obstruction may not be specific to early stages of PL. Bilateral hydronephrosis and abnormal renal function were detected at the second admission. The exact cause of CG is still unclear, but the obstruction of lymphatic vessels and veins caused by the compression of adipose tissue against the bladder may be the cause[8]. In this case, postoperative pathology confirmed CG of the intestinal epithelial type. At present, CG is generally divided into typical and intestinal epithelial types, with the latter showing a high tendency for malignancy. Therefore, cystoscopy is particularly important during early follow-up. Although intestinal metaplasia of the bladder is morphologically similar to that of the normal colon mucosa and other organs, the lack of expression of Hep, a marker of midgut epithelial metaplasia in bladder intestinal metaplasia, may indicate the presence of unique intestinal metaplasia pathway in the bladder[9].

There are no typical symptoms of PL. Imaging modalities such as CT may be the first choice for the diagnosis of PL, as opposed to surgery or biopsy[10]. Pelvic CT shows a symmetric increase in fat tissue around the bladder, and a change in bladder shape is important radiologically[10]. Studies have shown that "pear-shaped" and "banana-shaped" bladders are highly specific (100%) in predicting patients with PL, however, with a low sensitivity (40.6%)[10]. Zhang *et al*[11] analysed images from 32 cases of PL and found that the sensitivity and specificity for the diagnosis of PL by the bladder seminal vesicle angle were 62.5% and 100%, respectively, and that the optimal

threshold for diagnosis was 75°. In the previous literature, all patients diagnosed with PL had excretory cystograms or CT that demonstrated a bladder described as "pear-shaped", "banana-shaped", or "inverted teardrop-shaped" (Table 1). In our study, a similar imaging sign was detected, that is, the shape of the bladder was oval at the first visit, and it was pear-shaped on CT 6 mo after surgery. The bladder seminal vesicle angle increased from 49.5° to 89.5°. We speculate that in the early stages of PL, the bladder and sigmoid colorectum maintain a certain degree of tension and can withstand the compression from the excess fat, allowing the bladder to stay in a relatively normal shape. However, as the disease progresses, abnormal fat fills the gap between the lower ureter and the bottom of the bladder, narrowing the ureteral lumen, elongating the inner wall of the ureter, and then causing secondary hydronephrosis or stone formation. In our case, ureteroscopy was also performed because of hydronephrosis in both kidneys, and left lower ureteral stenosis was found along with ureteral calculi during the operation. In addition, all these anatomical changes make intraluminal surgery rather difficult. PL-induced hydronephrosis and ureteral dilatation are usually equal on both sides, but when there is asymmetry, the obstruction on the left side is usually more severe, possibly due to compression of the sigmoid colon[12], as in this case.

The cause of PL is unclear, but multiple studies have shown that PL may be related to obesity[13]. At present, a standard strategy for the treatment of PL needs to be recognized. Long-term oral antibiotics, hormone therapy, and external radiation therapy may be ineffective, and surgical intervention remains controversial[10]. Therefore, it is particularly important to diagnose PL early and slow its progression to hydronephrosis or even uraemia through conservative treatment.

Studies have shown that celecoxib can increase insulin sensitivity on subjects who are overweight or obese[14]. In a study by Hayashi *et al*[15], celecoxib inhibited the growth of tumours in obese mice with prostate cancer. In addition, the intestinal metaplasia in CG is similar to that in the colonic mucosa in terms of morphological changes and the expression of some markers[16]. Long-term oral celecoxib has been shown to prevent recurrence in patients with high-risk adenomas[17]. In addition, several studies have confirmed that COX-2 is overexpressed in CG[5,18]. Takizawa *et al*[6] reported the use of celecoxib to treat glandular cystitis, with certain effects. Our patient was diagnosed with refractory CG with PL and treatments including TUR-BT and long-term antibiotics turned out to be futile. We doubled the dose reported by Nae Takizawa (100 mg of celecoxib daily for treatment), which is also recommended in other inflammatory diseases such as adenoma[17] and bone arthritis[19]. In this patient, we used the CLSS questionnaire, which is a reliable tool for the comprehensive evaluation of LUTS in both sexes, to assess the patient's LUTS[20]. The patient showed a CLSS of 11 points at the first visit, and the CLSS stabilized at 8 points after comprehensive treatment, including oral celecoxib. Additionally, haematuria disappeared, and the serum creatinine level stabilized at 1.63 mg/dL (estimated glomerular filtration rate: 49 mL/min/1.73 m²). In addition, celecoxib improved the cystoscopy results, although subsequent CT scans showed PL with obstructive uropathy.

CONCLUSION

We report for the first time remission in a case of PL with refractory CG after TUR-BT and oral celecoxib. The aetiology of PL combined with intestinal CG is still unclear and needs further study, although it is generally related to chronic inflammation stimulation. COX-2 inhibitors may be a new strategy for the treatment of PL with refractory CG. The take-home messages from the current case include: (1) The early symptoms of PL are subtle and overlooked; (2) CT is very important in addition to cystoscopy; (3) PL patients need to be closely followed, because the condition may cause upper urinary tract obstruction or even renal failure as it progresses, and the surgical treatment effect leaves much to be desired; and (4) COX-2 inhibitors may be a new strategy for the treatment of PL with refractory CG.

Table 1 Pelvic lipomatosis with cystitis glandularis reported in the previous literature

Case (No.)	Age (yr)	Sex	Symptoms	Radiographic findings	Treatment	Outcome	Ref.
1	48	Male	Nocturia	Banana-shaped	Surgical removal of fat	Subjective symptoms improved, radiographic findings not changed	Fogg <i>et al</i> [1], 1968
2	35	Male	Perineal	Teardrop-shaped	Radical cyst prostatectomy	Radiographic findings no recurrence of tumour	Heyns <i>et al</i> [3], 1991
3	47	Male	LUTS	Banana-shaped	Surgical treatment using a bladder-sparing technique	Hydronephrosis not changed	Ge <i>et al</i> [7], 2019
4	49	Male	LUTS	Pear-shaped	TUR-BT and oral administration of celecoxib	Subjective symptoms improved, cystitis glandularis improved, radiographic findings not changed	Our case

LUTS: Lower urinary tract symptoms; TUR-BT: Transurethral resection of bladder tumour.

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