

Pelvic Lipomatosis Associated with Cystitis Cystica and Chronic Metaplasia of the Urinary Bladder: a case report

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Pelvic lipomatosis is a rare disease by excessive proliferation of the mature fat in the pelvic extraperitoneal space. Cystitis cystica, often occurring concurrently with pelvic lipomatosis, is a rare proliferative inflammatory disease of the bladder. The diagnosis of pelvic lipomatosis is established on radiographic examination, not on pathology. We report a case of pelvic lipomatosis associated with cystitis cystica and chronic metaplasia of the urinary bladder.

Key words: *Cystitis cystica; Lipomatosis*

Pelvic lipomatosis is a rare disease of unknown etiology presenting with excessive proliferation of the mature fat in the pelvic cavity. The proliferating adipose tissue may compress the pelvic viscera, including the pelvic portion of the ureters, rectosigmoid colon and vessels, in varying degrees. Therefore the presenting symptoms usually vary and suggest nonspecific diseases referable to the pelvis or urinary tract. There may be no symptoms in pelvic lipomatosis and is usually discovered incidentally. But occasionally, marked bilateral ureteral obstruction may lead to the development of uremia.

Pelvic lipomatosis has been associated with various yet often unrelated disorders including: cystitis cystica, cystitis glandularis, adenocarcinoma of the bladder, chronic urinary tract infection, hypertension, recurrent superficial thrombophlebitis [1], constipation, vesicoureteral reflux, retroperitoneal fibrosis, nontropical chyluria [2], and the Proteus syndrome [3]. Therefore early diagnosis and early treatment of associated disorders before late sequelae develop are important. Here, we reported a case of pelvic lipomatosis associated with cystitis cystica proved by cystoscopic biopsy.

CASE REPORT

A 25-year-old young man was admitted due to progressive frequency and left inguinal pain in recent 2 days. He had suffered from recurrent frequency, dysuria, nocturia, painless hematuria, lower abdominal soreness and bilateral flank soreness since 3 years ago.

Physical examination revealed 180.6 cm in body height and 68 kg in body weight. There was no evidence of bleeding tendency, abdominal tenderness, costophrenic angle knocking pain, fever, hypertension, hypotension, hemihypertrophy, macrodactyly, various subcutaneous masses or scoliosis. But left spermatic cord tenderness was noted. Urine routine showed proteinuria, hematuria and pyuria.

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1a



1b

Figure 1. Intravenous urography (IVU): **a.** At-60-minute spot film shows severe bilateral hydronephrosis and hydroureters with tapering of bilateral lower ureters and marked elevation of the bladder base. **b.** U-bladder filling-view discloses diffuse radiolucent area in the pelvic cavity. The bladder simulates an inverted pear.

The plain film of the abdomen showed radiolucent area in the pelvis. The intravenous urography (IVU) demonstrated severe bilateral hydronephrosis and hydroureters with tapering of bilateral lower ureters and marked elevation of the bladder base (Fig. 1a). The bladder had a strikingly abnormal configuration, simulated an inverted pear (Fig. 1b). There was also straightening and elevation of the rectosigmoid colon on a barium enema study (Fig. 2). CT revealed abundant fatty tissue with increased vascularity in the pelvic space, bladder deformity and mild rectal compression. Small cystic lesions in the thickened bladder wall secondary to inflammation, edema and proliferative changes were also detected on CT image (Fig. 3). The magnetic resonance image (MRI) simulated CT findings. Thickening of the urinary bladder wall, dilated bilateral lower ureters and compressed rectum were, however, more prominent than CT due to better tissue-contrast in MR image (Fig. 4).

Cystoscopy revealed polyps in trigone of the urinary bladder and prostatic urethra. Transurethral multiple biopsies of the urinary bladder in body, neck, and posterior trigone portions were performed. The pathological report was cystitis cystica (Fig. 5a) with intestinal epithelium metaplasia noted in the mucosa and submucosa of the urinary bladder (Fig. 5b).

After the diagnosis was confirmed, bilateral percutaneous nephrostomies were performed; bilateral double J stents were also inserted antegradely. After the symptoms were released, the pigtail catheters of



Figure 2. Barium enema study demonstrates straightening and elevation of the rectosigmoid colon.

bilateral percutaneous nephrostomies were removed. The patient was then discharged and regularly followed up at our out-patient department.

DISCUSSION

Pelvic lipomatosis is nonmalignant overgrowth of adipose tissue with minimal fibrotic and inflammatory components compressing structures within the pelvis. It occurs most frequently in a specific group of patients: middle age (25-60 years old); usually over-

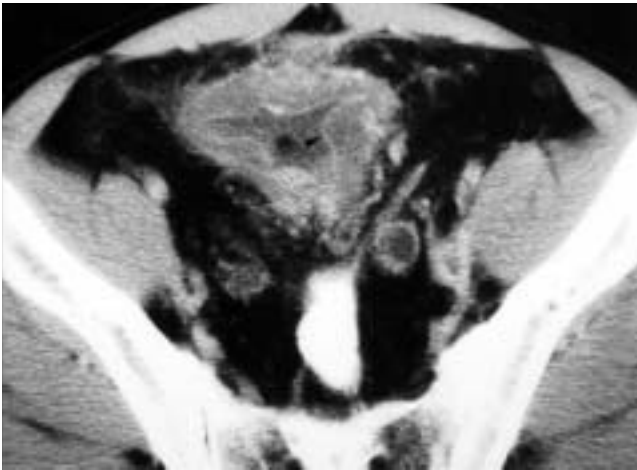


Figure 3. Contrast-enhanced CT scan of the pelvis reveals abundant fatty tissue with increased vascularity. The bladder wall is markedly thickening due to inflammation and edema. Cystic lesions are noted in thickened bladder wall (►). Dilated bilateral lower ureters and mild rectal compression are also seen.

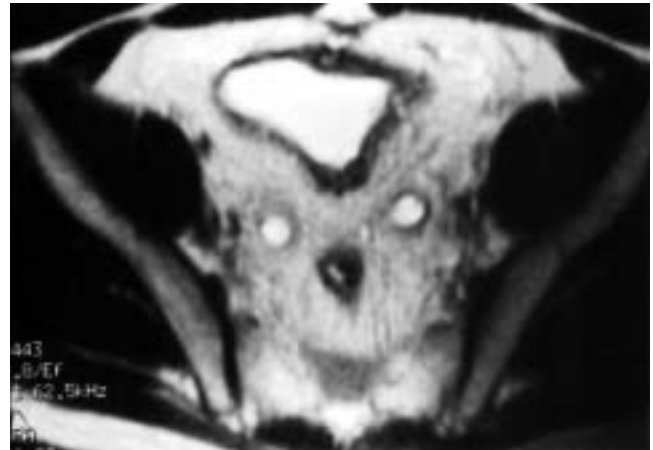
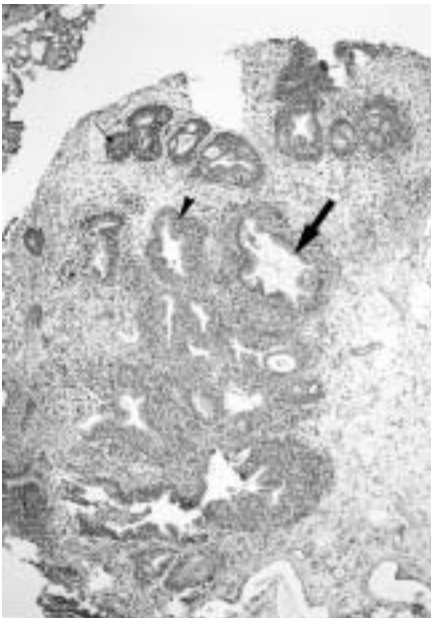
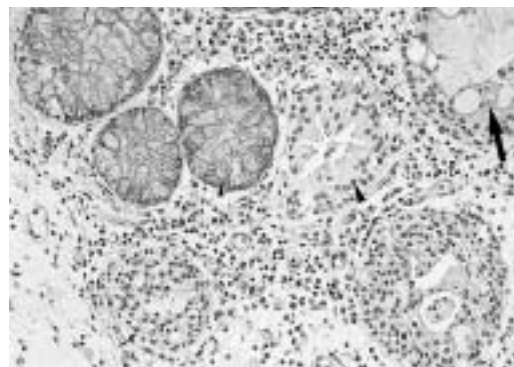


Figure 4. Fast spin echo T2-weighted MR image (27443/95 msec/0.50) (TR / TE / excitations) on axial section of the pelvis demonstrates similar findings as **Figure 3**: thickening of the urinary bladder wall, dilated bilateral lower ureters and compressed rectum are, however, more prominent than CT due to better tissue-contrast in MR image.



5a



5b

Figure 5. Histological examination of the urinary bladder mucosa reveals chronic inflammation with morphology of von Brunn's nests (→), cystitis cystica (►), cystitis glandularis (⇨), and chronic intestinal metaplasia (▶). (a. X40, HE stain), (b. X200, HE stain)

weight but not obesity, and man predominant (10:1) [4]. Clinical symptoms are nonspecific and include urinary frequency, urinary tract infection, dysuria, hematuria, fever, constipation, and abdominal or low back pain. Therefore pelvic lipomatosis is often found incidentally on radiography.

The diagnosis is suggested by radiological triad of hyperlucency of the pelvis on plain abdomen film,

“hot air balloon” appearance of the bladder on IVU [5] and a rigid and ascended rectosigmoid colon on barium enema. The final diagnosis of pelvic lipomatosis is confirmed by CT and MRI findings. The ultrasonography might show hyperechoic masses and bladder deformity. But due to marked echogenicity associated with fat and bowel gas, a specific diagnosis usually cannot be made. Cystourethrography reveals

an elevated pear-shaped bladder and an elongated and anteriorly displaced prostatic urethra [6]. Although multiplanar MRI is able to detail fatty masses and provides delineation of cephalad displacement of the bladder base, elongation of the bladder neck and posterior urethra, and elevation of the prostate gland, it offers little benefit over CT except for showing characteristic medial and superior displacement of the seminal vesicles and fatty tissue separating the prostate gland from the rectum [7]. Pelvic angiography is helpful to distinguish inflammatory disease from neoplastic disease. Inflammatory vessels are smaller (less than 0.3 mm in diameter) and the venous phase appears later than in malignancy. An increased vascular supply is always noted in pelvic lipomatosis, but the characteristic larger vessels, rapid venous filling, tumor stain, displacement, and amputation associated with malignancy are not found. It is emphasized that the differentiation of pelvic lipomatosis from malignant neoplasm without laparotomy depends on demonstration of the typical radiolucency surrounding the bladder and rectum. But if malignancy is highly suspected, exploratory laparotomy may be necessary [8].

An increased incidence of proliferative cystitis such as cystitis cystica or cystitis glandularis in associated with pelvic lipomatosis has been noted. The cause of these associated proliferative changes, however, remains unknown [8]. Cystitis cystica is a nonspecific inflammatory process of the urinary bladder wall. It is commonly seen with von Brunn's nests and cystitis glandularis in the inflamed bladder or around the stalk of papillary tumors and the margins of sessile or invasive tumors. Von Brunn's nests, cystitis cystica, and cystitis glandularis are three related condition. Von Brunn's nests are formed by buds or sprouts of transitional epithelium that grow down from the surface into the underlying lamina propria and later become surrounded by condensed layer of connective tissue that eventually separated them from the overlying cells. Central cavitation of the nests or infolding of the surface epithelium, with the formation of crypts that become obstructed, will lead to the formation of cystitis cystica. In the fully formed cysts there is a lining of flattened or low cuboid cells, and if the cysts are of sufficient size, the mucosal surface becomes irregular and has a granular appearance. Cystitis glandularis is produced when the cells lining the cysts differentiate into columnar epithelium. Typical goblet cells are sometimes present, and the epithelium resembles intestinal mucosa named intestinal metaplasia. It is widely regarded as premalignant and may evolve into adenocarcinoma of the urinary bladder. There is increased risk of adenocarcinoma if more extensive

intestinal metaplasia develops in the bladder wall [9].

These chronic proliferative changes of the bladder are demonstrated as multiple small round filling defects on IVU [10], mucosal elevations on ultrasound and cystic lesions may be seen within the thickened bladder wall on CT [11] and MRI image. Sometimes a single defect may mimic transitional cell carcinoma [12] and the final diagnosis is based on histology.

The prognosis of pelvic lipomatosis depends on various factors including onset of the disease, if there is association with other disorders, and development of sequelae. There is a higher risk of progressive ureteral obstruction in young, stocky or obese men with vague pelvic symptoms. While pelvic lipomatosis in men older than 60 years old usually discovered during evaluation of other problems, often prostatism, might have no serious sequelae and no significant progression of disease [8]. Some patients with pelvic lipomatosis might develop uremic encephalopathy if they are loss of follow-up. Therefore regular follow-up and an early intervention before irreversible changes occur are important.

In conclusion, pelvic lipomatosis is usually associated with cystitis cystica and chronic metaplasia. Early diagnosis and early treatment of these associated disorders before late sequelae develop can be achieved through careful radiological evaluation. ◆

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骨盆腔脂肪增多症合併囊狀膀胱炎跟慢性膀胱化生：病例報告

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骨盆腔脂肪增多症是一種成熟的脂肪在骨盆腔中過度增生的罕見疾病。囊狀膀胱炎是一種罕見的膀胱增生性發炎性疾病，常與骨盆腔脂肪增多症合併發生。骨盆腔脂肪增多症的診斷主要是建立在影像學的檢查上而非病理變化。我們報告一個骨盆腔脂肪增多症合併囊狀膀胱炎及慢性膀胱化生的病例。

關鍵詞：骨盆腔脂肪增多症；囊狀膀胱炎