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Case Report

Surgery Section

Pelvic Lipomatosis in a Young Male: A Case Report

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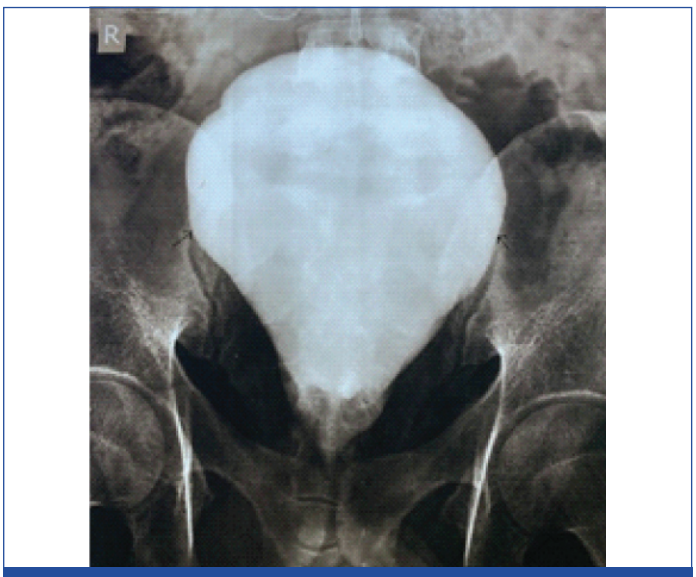
ABSTRACT

Pelvic Lipomatosis (PL) is a rare benign disease of unknown aetiology characterised by intrapelvic overgrowth of mature fatty tissue. Although non malignant, the adipose tissue is infiltrative and causes compression of the pelvic structures-the genito urinary tract, lower gastrointestinal tract or the vascular system leading to a broad range of symptoms. With limited cases of PL reported in the literature there are no clear-cut guidelines for its management and both conservative and surgical modalities have been described with varying results. This report describes a case of PL in a young 34-year-old male which was associated with bilateral hydroureteronephrosis and was well managed conservatively. PL is also known to be associated with complications like hypertension, severe upper tract obstruction, renal failure and rarely pulmonary thromboembolism, portal vein thrombosis and adenocarcinoma bladder, thus commanding early diagnosis of the disease and long-term follow-up in all the patients.

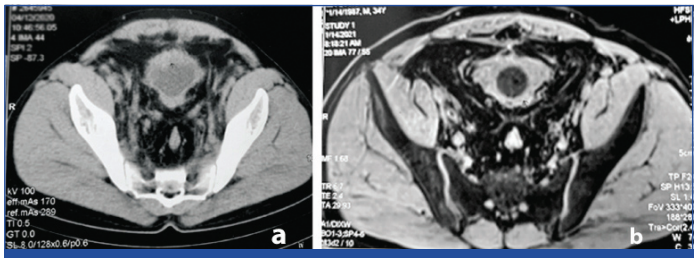
Keywords: Adipose tissue, Bilateral hydroureteronephrosis, Thromboembolism

CASE REPORT

A 34-year-old male presented to the department with complaints of dysuria, increased frequency, straining to void and poor stream since four years. He had history of seven pack years of smoking with no other relevant past medical history. There was no history of altered bowel habits or erectile dysfunction. Physical examination and digital rectal examination were unremarkable and the body mass index of the patient was 21 kg/m^2. Ultrasonogram revealed Grade 2 hydronephrosis of left kidney, dilated left ureter with huge postvoidal residual urine (334cc) and a polyp in urinary bladder. Uroflowmetry of the patient showed decreased maximum flow rate (Qmax) of 8 mL/sec. On Micturating Cystourethrogram no obvious abnormality was seen except 'pear' shaped urinary bladder [Table/Fig-1]. The routine blood tests were normal and the urine was negative for malignant cells. Computerised Tomography (CT) urogram of the patient revealed diffusely thickened urinary bladder wall with soft tissue stranding into perivesical fat, mural thickening with mild narrowing of both lower ureters causing bilateral hydroureteronephrosis [Table/Fig-2a]. Magnetic Resonance Imaging (MRI) of pelvis also confirmed diffuse thickening of pelvic preperitoneal fat planes [Table/Fig-2b]. This was followed by Cystourethroscopy which revealed mucosal irregularities with a 1×1.5 cm polyp at the left side of bladder neck.

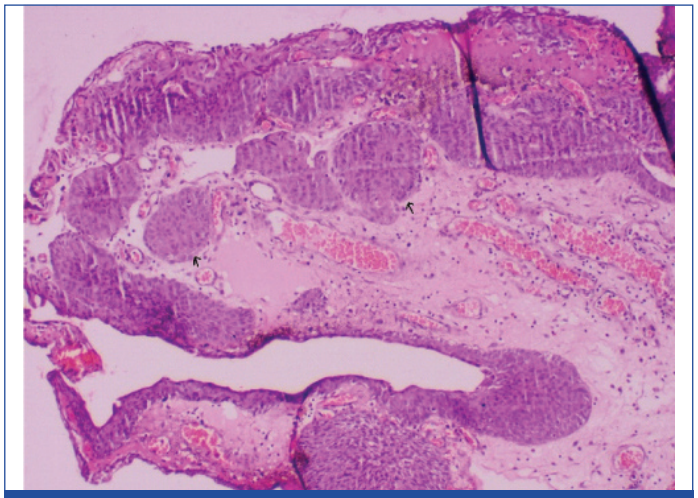


[Table/Fig-1]: Micturating cystourethrogram showing 'pear' shaped urinary bladder.



[Table/Fig-2]: a) Computed tomography of the pelvis showing diffuse thickened urinary bladder wall; b) MRI of the pelvis showing diffuse thickening of pelvic preperitoneal fat planes and urinary bladder wall.

On the basis of above investigations the diagnosis of urinary bladder space occupying lesion with PL was made and a tissue biopsy was planned. Intraoperatively, a mass with a solid base with papillary projections was seen at the neck and trigone of urinary bladder extending to the prostatic urethra. Transurethral Resection of Bladder Tumour (TURBT) was done and the tissue was sent for histopathological examination. The microscopic findings included normal transitional epithelium along with cyst like structure within the stromal tissue lined by transitional cells, solid nest of benign urothelial cells with regular contours in the stroma and no granuloma or atypia in the muscularis propria. All the above features on the histopathology pointed towards a benign pathology, Cystitis cystica [Table/Fig-3].



[Table/Fig-3]: Histopathology examination of resected urinary bladder lesion showing cyst like structure within the stromal tissue lined by transitional cells, suggestive of cystitis cystica.

A conservative approach with regular follow-up was planned. At three months follow-up, USG showed mild hydronephrosis on the left side with PVR-75 mL and there was symptomatic improvement noticed. After 10 months of follow-up, the symptoms of the patient are still under control with normal renal parameters.

DISCUSSION

The PL was first described by Engels in 1959 [1]. It is a rare disease characterised by non malignant overgrowth of normal adipose tissue in the perivesical and perirectal spaces. PL is known to be most prevalent in men and individuals with dark-skinned phenotype, most frequently at the third or fourth decade of life. The exact aetiology of PL remains unknown. It was initially thought to result from chronic lower urinary tract infection with the overproduction of fat as a part of an inflammatory response [1,2]. Some authors have raised the hypothesis that this disease is a manifestation of generalised obesity or an underlying endocrine dysfunction like diabetes mellitus, Cushing's disease or hyperthyroidism [3-5]. Venous stasis has also been proposed as a possible cause of PL based on the remarkable vascularity associated with the pelvic fat [6].

The clinical presentation of PL depends on the compressed tissues and varies from dysuria, nocturia, haematuria, constipation, tenesmus, rectal bleeding to oedema of lower extremities. Physical examination may reveal pain on abdominal palpation, a palpable mass in the hypogastric region, elevation of prostate at digital rectal examination or lower limb oedema. Definitive diagnosis is made with the help of imaging modalities which demonstrate deposition of fatty tissues causing compression of adjacent structures, resulting in morphological deformities. Hypertension, renal failure and rarely pulmonary thromboembolism, portal vein thrombosis and bladder adenocarcinoma are the complications that have been described in the literature [7,8]. In the present case, bilateral hydroureteronephrosis was detected probably because of bladder compression.

Both the conservative treatment approach with regular monitoring of renal function in patients with less aggressive disease and surgical modalities in patients with more aggressive disease have been described in literature with varying results [9-11]. Various surgical procedures like ileal conduit with or without simple cystectomy, cutaneous ureterostomy and bilateral percutaneous nephrostomies, to treat bilateral hydroureteronephrosis have been described in the past. In 2012, Gupta SK et al., reported a case of PL in a 40-year-old male who was managed successfully by simple cystectomy and ileal conduit. The clinical and radiological presentation in this case was very similar to our case with both the patients having lower urinary tract symptoms, low flow rate on uroflowmetry, classical 'pear' shaped bladder on micturating cystourethrogram and CT and MRI findings supporting the diagnosis [9]. In 2019, Ge L et al., reported a series of 8 patients with PL who underwent laparoscopic bladder fat extirpation and B/L ureteric reimplantation with a median follow-up of 48.5 months and only one patient was reported to have recurrence at 49 months [10]. Another recent study of 5 patients with PL who underwent robotic-assisted wide bladder fat extirpation and B/L ureteric reimplantation with DJ stenting was reported with short-term success [11]. The various surgical challenges that have been reported in PL are lack of surgical planes, presence of tough, thick, adherent, and hypervascular fat planes that bleed easily on manipulation, difficulty in separating the ureter from adherent tough fat and bladder mucosa buried underneath the thick fat planes, making anastomosis difficult [11]. Mo LC et al., in a recent report has shown good outcome in a patient of PL with cystitis glandularis with COX-2 inhibitor and TURBT [12]. However, PL management modalities lack large scale trials with long-term efficacies with only individual reports or series available with limited follow-up. In our case, the patient was young with normal renal parameters and had symptomatic improvement post TURBT, so, a conservative approach with regular follow-up was chosen to determine the progression of disease and to prevent any further complications. Additionally, the patient was explained about the need for long-term surveillance for adenocarcinoma bladder.

CONCLUSION(S)

Although surgical procedures like cystectomy, pelvic fat mass extirpation form one of the main treatment options for the management of PL but selected cases like ours can be managed with endourological procedure, provided the patient is willing for long-term follow-up.

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