International Journal of Current Advanced Research
ISSN: O: 2319-6475, ISSN: P: 2319-6505, Impact Factor: 6.614
Available Online at www.journalijcar.org
Volume 10; Issue 08 (A); August 2021; Page No.24897-24900
DOI: http://dx.doi.org/10.24327/ijcar.2021.4964.24900

Case Report

PELVIC LIPOMATOSIS- A BLADDER PRESERVING APPROACH – A CASE REPORT

Sinha Sujeet., Palaniyandi Velmurgan and Natarajan Kumaresan*

Department of Urology Sri Ramachandra Institute of Higher Education and Research (SRIHER), Chennai, Tamilnadu, India

ARTICLE INFO

Article History:
Received 4th May, 2021
Received in revised form 25th June, 2021
Accepted 18th July, 2021
Published online 28th August, 2021

Key words:
Pelvic Lipomatosis

ABSTRACT

Pelvic lipomatosis is a rare condition of unknown cause characterized by diffuse exuberant pelvic overgrowth of non-malignant but infiltrative adipose tissue in the perivesical and perirectal space[1], first reported in 1959 by Engles[2]. Around 70% of patients are found to be associated with proliferative cystitis, such as cystitis cystica and cystitis glandularis[1,3]. CT scans are found to be the most competent and crucial imaging[4]. A pear–shaped bladder is a distinctive feature on CT urogram in pelvic lipomatosis[5]. The various treatment options available are long–term antibiotics, transurethral resection of the lesions, cystectomy and urinary diversion[6]. The present report describes a case of 45–year–old male patient with pelvic lipomatosis presented with voiding symptoms, persistent pelvic pain and poor quality of life. A bladder–sparing excision of the pelvic lipomatosis mass was performed, thereby avoiding the need for urinary diversion.

INTRODUCTION

Pelvic Lipomatosis is a rare disease more common in middle aged man. It requires a high degree of clinical suspicion and radiological features. Cystoscopy should be done during evaluation to rule out concomitant Adenocarcinoma of Urinary bladder. Patient can be managed conservatively or excision of mass with or without Urinary diversion, depending upon clinical and radiological features. Our Patient was managed with excision of mass without urinary diversion with significant symptomatic improvement and post op cystogram showing significant improvement in bladder size capacity. Patient is on regular follow up.

Case Presentation

A 45-year-old, obese, hypertensive, diabetic patient presented with increased frequency, straining to void with terminal dribbling and burning micturition for one month. History of hematuria, passage of mucus on and off present. Physical examination suggestive of vague, non tender, intra-abdominal lump arising from pelvis reaching midway between pubic symphysis and umbilicus, lower border not palpable with limited mobility. External genitalia and digital rectal examinations were normal.

Investigation

Ultrasound Abdomen suggestive of vertically distended pear shaped bladder, elongated bladder neck, bilateral hydro ureteronephrosis with ureters dilated up to mid third of their length.

Corresponding author:* **Natarajan Kumaresan
Department of Urology Sri Ramachandra Institute of Higher Education and Research (SRIHER), Chennai, Tamilnadu, India

Computed Tomographic urogram showed increased deposition of fat in pelvis, elongated pear shaped urinary bladder with Bilateral hydro ureteronephrosis till distal third [Figure 1,2,3]. Laboratory investigations including serum creatinine(0.8 mg/decilitre), hemoglobin (14.3 grams/decilitre) were within normal limits.

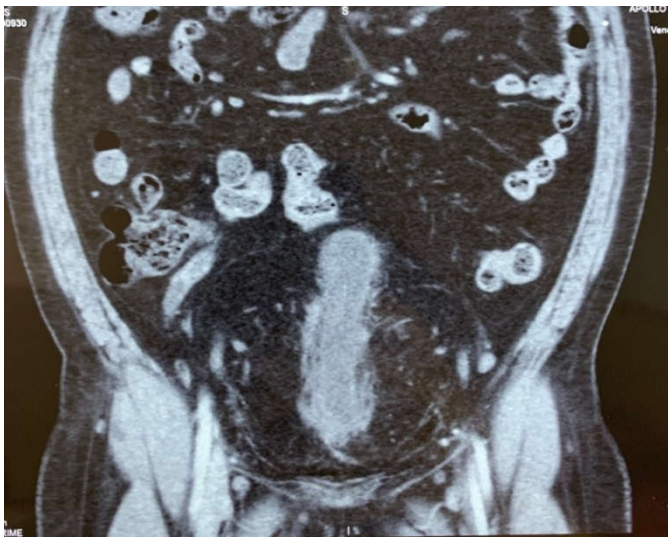

Figure 1 Computed Tomography Abdomen showing pear shaped elongated bladder in a coronal section with surrounding locculated fat (pre operative image)

Differential Diagnosis

Pelvic liopma – pelvic lipoma was ruled out based on radiological study because liopma usually are unilateral and

well defined, where as in out patient it was uniformly distributed and was involvingbbladder from all sides.

Liposarcoma – Liposarcoma was ruled out based on radiology because liposarcoma usually infiltrate surrounding organs and can infiltrate bladder, where as in out patient lesion was confined to pelvis and was not infiltrating any organs just surrounding and compressing bladder all around.

Treatment

Patient underwent Cystoscopy + Left double J stenting with bladder biopsy. Per op– Right ureteric orifice was not visible. Bullous lesion on trigon and right lateral wall with Grade 1 bladder trabeculation. Bladder capacity was 200 milllitres.

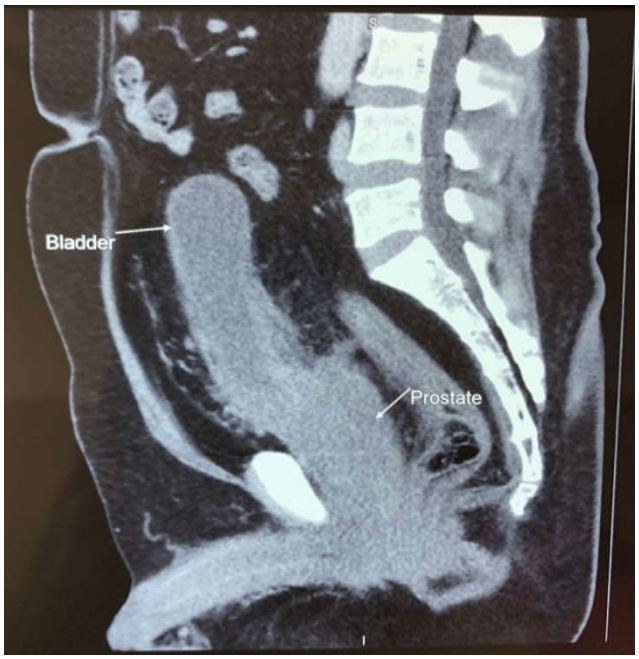

Figure 2 Computed Tomography Abdomen showing pear shaped elongated bladder in a sagittal view with surrounding fat (pre operative image)

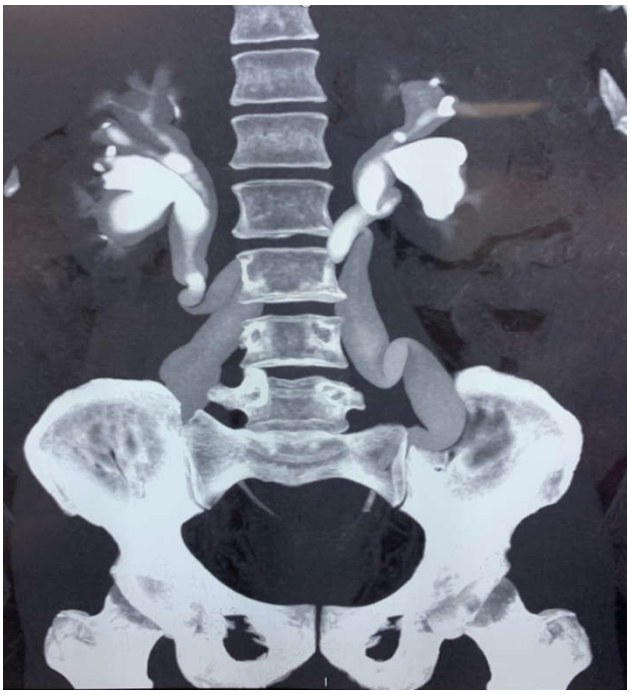

Figure 3 Computed Tomography Abdomen with 3D reconstruction of kidney ureter bladder region showing bilateral hydro ureteronephrosis (pre operative image)

Bladder biopsy histopathology was suggestive of Cystitis cystica glandularis with intestinal metaplasia. Patient then underwent Laparotomy with excision of perivesical and pelvic fat. The bladder was bivalve and mobilized, the pelvic and perivesical fat was excised completely.

Outcome and Follow-Up

Post operatively patient recovered well, a marked improvement of the patient's symptoms was noted with bladder capacity reaching approximately 400 millilitres. Post operative Computed tomography urogram and cystogram were done to compare pre and post operative status and capacity of the bladder, both showed significant improvement [Figure 4,5].

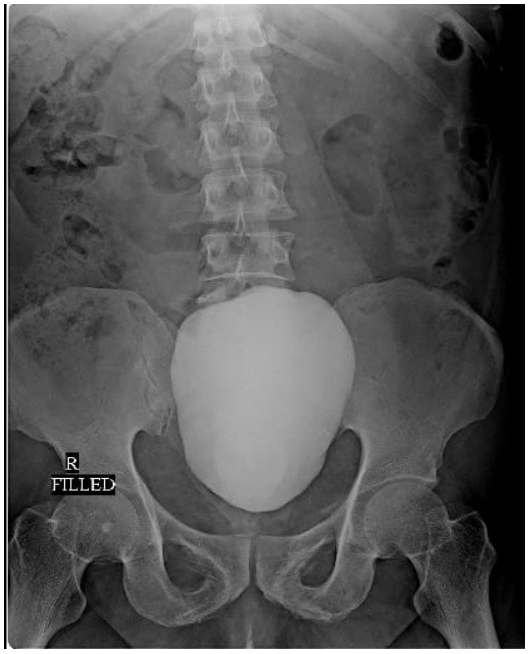

Figure 4 Cystogram study showing well distended bladder in pelvis with well-maintained contour (post operative image)

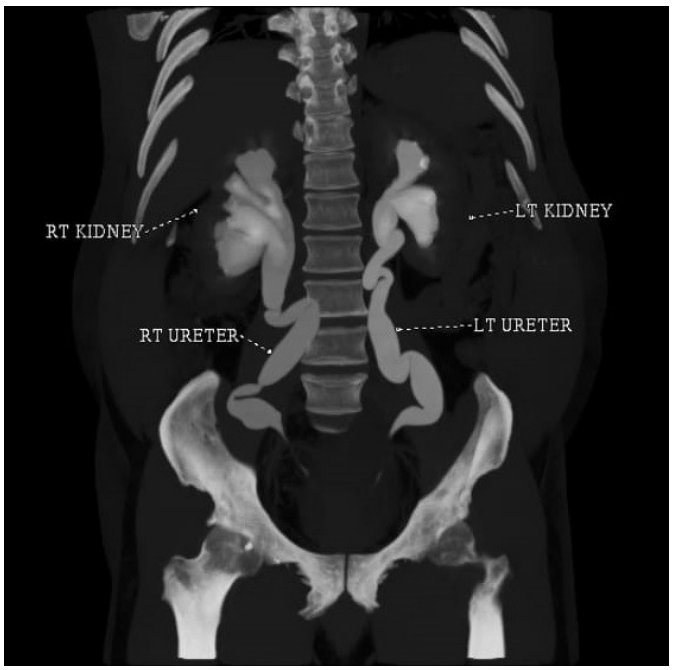

Figure 5 Computed Tomography Abdomen with 3D reconstruction of kidney ureter bladder region showing residual bilateral hydro ureteronephrosis, reduction noted as compared to pre operative image (post operative image)

DISCUSSION

Pelvic lipomatosis is a rare condition characterized by diffuse exuberant pelvic overgrowth of non-malignant but infiltrative adipose tissue in the perivesical and perirectal space [1]. The disease, first reported by Engels in 1959 [2], is characterized by overgrowth of non capsulated, non malignant but infiltrative adipose tissue. Fogg and Smyth first coined the term pelvic lipomatosis in 1968 [7]

The incidence of pelvic lipomatosis is 0.6 to 1.7 per 100,000 hospital admissions [1]. The mean age of presentation is 48 years [1]. There is a strong male predominance, with a male-to-female ratio of 1.8:1 [1]. There is also a racial disparity: 67% of African Americans have pelvic lipomatosis compared to 33% of Caucasian patients [1].

Histologic examination reveals normal, mature adipose tissue. Occasionally, mild fibrosis and inflammatory cells can be noted. Pelvic lipomatosis is usually symmetrically distributed and confined to the pelvis, however, retroperitoneal involvement may sometimes occur [7]

The etiology of this disorder is unknown, but it is suggested that obesity plays a role, and it has been found in half of the patients afflicted with obesity [8,9]. A possible genetic etiology is suggested by abnormality in the chromatin-regulatinghigh motility group A (HMGA) proteins [10]. High motility group A2 (HMGA2) overexpression and high motility group A1 (HMGA1) under expression are associated with increased fatty tissue deposits in murine models [11-13].

The clinically patient may experience symptoms based on manifestations from the extrinsic compression of the structures comprising the urinary system, the lower intestinal tract and the vascular system. Clinical symptoms that are associated with the advanced stage of the disease, as well as physical findings are usually nonspecific and unremarkable, Suprapubic pain or lumps, backache, flank pain, or perineal discomfort may be other clinical manifestations [14]. Associated hypertension is common, seen in 35 to 74% of patients [3]. Hydronephrosis develops in at least 1/3 of the patients evaluated [3]. It often presents with bilateral hydronephrosis in 19% of patients. Hydronephrosis occurs due to ureter's distal encasement by lipomatosis lesions

Renal failure occurs in 6% of all reported cases. This small incidence of renal failure is attributed to a gradual, progressive course of pelvic lipomatosis, and a high grade of obstruction [15]. A significant complication of pelvic lipomatosis is thromboembolism, which can be expressed by either deep venous thrombosis or pulmonary embolism (7%). Venous stasis is due to the compression of iliac veins or vena cava [15-17]. The possible late complications are obstructive renal failure, proliferative cystitis [3] and bladder adenocarcinoma [3,5]

On general physical examination, patients may have lower limbs edema and arterial hypertension. On abdominal examination patient may have pain on abdominal palpation, a palpable mass in the hypogastric region, occasionally urinary retention, elevated prostate on digital rectal examination, [14,18].

The condition is diagnosed based on a large index of suspicious clinical and radiological findings. The first radiological indicator is marked translucency in the perivesical area on plain X-ray kidney, ureter, bladder(KUB) film [19]. Excretory urography demonstrates hydroureteronephrosis and deviation of ureters to the medial in 80% or to the lateral in 20% of patients. the bladder characteristically assumes a pear or gourd shape, with an elevated bladder base [20]. The lack of prostatic indentation at the base of the bladder is an important differential point in the exclusion of prostatic enlargement as the cause of bladder elevation [20]. On pelvic and abdominal Computed Tomography, the bladder and rectosigmoid are surrounded and displaced by homogenous tissue with low attenuation (-40 to -100 Hounsefield units), signifying fat content. The Computed Tomography establishes a diagnosis of pelvic lipomatosis [21,22]. An Magnetic Resonance imaging can also be used for diagnosis, as it permits the characterization of fat planes, and it provides the delineation of an elevated bladder base and the elongation of a posterior urethra [23] The fat tissue in the pelvis and retroperitoneum is extensive, homogenous, nonencapsulated, and usually bilateral and symmetrically distributed Evaluation by cystoscopy and the biopsy of any suspicious lesion should be taken because of an association with adenocarcinoma of the bladder [3]. Cystoscopy shows abnormalities in approximately 75% of patients and most often findings are of cystitis glandularis and cystitis cystica [3]

Differential diagnosis for lesions mostly composed of the fat in the retroperitoneum and pelvis would include pelvic lipomatosis, lipoma and liposarcoma. Lipoma cannot be distinguished radiographically from pelvic lipomatosis but they are usually unilateral, encapsulated, and less extensive comparing to pelvic lipomatosis, which is usually symmetrical and bilateral. Fatty masses that remain stable in size, together with long time history of the disease, can help in excluding malignant disease [24]

Pelvic lipomatosis can be managed conservatively, till it isn't complicated by obstruction of the urinary tract, cystitis glandularis, or adenocarcinoma.

Antibiotics, staroids, and radiation therapy have been tried but not much success reported [1]. Urinary diversion, in the form of nephrostomy tube, ileal conduit, or vesicostomy, are options to relieve urinary obstruction associated with the condition [7]. After discussing various treatment options, the patient opted for a bladder–preserving procedure due to his age and intention to preserve his quality of life.

Due to the consequences of the chronic obstruction, the chronic vesical wall irritation or even compression of the lymph chains caused by the deposited fat tissue may explain the common finding of proliferative processes involving the bladder mucosa such as cystic and glandular cystitis.

In spite of being considered as a benign lesion, there is remote possibility of development of vesical adenocarcinoma, justifying the periodic follow-up with cystoscopy, urinary cytology and biopsy of suspicious lesions(3).

Take Home Message

Pelvic Lipomatosis is rare disease but high degree of clinical suspicion and radiological features helps in diagnosis.

Cystoscopy to rule out concomitant Adenocarcinoma of Urinary bladder is necessary.

Patient can be managed conservatively, excision of mass with or without urinary diversion with regular follow up with cystoscopy.

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How to cite this article:

Sinha Sujeet *et al* (2021) 'Pelvic Lipomatosis- A Bladder Preserving Approach – A Case Report', *International Journal of Current Advanced Research,* 10(08), pp. 24897-24900. DOI: http://dx.doi.org/10.24327/ijcar.2021.4964.24900

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