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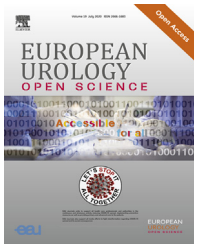

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Reconstructive Urology

Robotic Management of Pelvic Lipomatosis—Experience with Difficulties Encountered and the Techniques to Successful Outcomes

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Abstract

Background: Pelvic lipomatosis (PL) is a rare condition characterized by diffuse pelvic overgrowth of nonmalignant but infiltrative adipose tissue in perivesical and perirectal space.
Objective: To share our robotic experience and difficulties encountered and suggested techniques to overcome them successfully. It is the first series from India.
Design, setting, and participants: A prospective observational study was conducted. All consecutive patients diagnosed with PL between 2016 and 2019 underwent robotic-assisted wide bladder fat extirpation and bilateral ureteral reimplantation (extravesical refluxing type) with double J stenting and were evaluated prospectively.
Outcome measurements and statistical analysis: Demographics, serum creatinine level, radiographic features, postoperative complications, and patient-reported outcomes were evaluated. Continuous variables are presented as median and range, as the sample size is very small.
Results and limitations: We encountered a total of five patients with PL. The median console time was 126 (range 120–130) min, with a median estimated blood loss of 120 (range 100–150) ml. Postoperative complications were Clavien-Dindo grade I in three cases, and the median hospital stay was 2 d. Distal ureteric margins showed subepithelial edema with submucosal fibrosis, and bladder fat biopsies were reported as adipose tissue with hemorrhage. At initial 3-mo and annual follow-ups, all patients had normal serum creatinine and there was no evidence of disease recurrence. Limitations of our study are the very small sample size (a low incidence of PL) and short follow-up time period (the question of how long the surgical effect will be sustained due to abnormal proliferation of fat cells remains unanswered).
Conclusions: Robotic management of wide bladder fat extirpation and bilateral ureteral reimplantation with double J stenting has a good success rate and good outcome in PL.

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Patient summary: Pelvic lipomatosis is a very rare condition. Robotic management in treating the condition has good outcome for the patient. Here, we have discussed the difficulties encountered in treating the condition and the techniques used to overcome them.

1. Introduction

Pelvic lipomatosis (PL) is a rare condition characterized by diffuse pelvic overgrowth of nonmalignant but infiltrative adipose tissue in perivesical and perirectal space [1]. It was first reported by Engels [2] in 1959 in five patients. The incidence rate is 0.6–1.7/100 000 hospital admissions. So far, only <150 cases have been reported worldwide, with a largest single-center series of 35 cases over a period of 18 yr in the Medical College of Virginia Hospitals [3]. It presents with a broad range of symptoms caused by compression of pelvic structures [4]. Differential diagnosis includes pelvic teratoma, retroperitoneal fibrosis, pelvic lipoma, and liposarcoma based on the features on imaging [5]. The etiology being obscure, various treatment options, such as dietary modification, antibiotics, steroids, and radiotherapy, had been proposed with limited efficacy. Carpenter [6] proposed surgical management based on age. In the older group (>50 yr), conservative surgical procedures, such as ureteral stenting or transurethral resection of the prostate, was suggested to provide symptomatic relief. However, in the stocky and obese younger age group (<50 yr), the disease is more aggressive and progressive, leading to bilateral (B/L) ureteral obstruction; hence, urinary diversion procedures were performed. It is unfair to offer such morbid procedures for a benign disease in such young patients. With the evolution of technology over the years, minimal-access bladder-sparing surgeries have been performed with a good success rate. Ge et al [7] published a series of eight cases (second largest series over a period of eight years) who underwent laparoscopic procedure with good outcomes. We present our experience of robotic-assisted bladder-sparing surgery with good short-term outcomes, and it is the first series from India.

2. Patients and methods

2.1. Aim of the study

The objective of this study is to share our robotic experience in treating PL, the difficulties encountered during surgery, and the techniques suggested to overcome these difficulties successfully.

2.2. Study design

This is a prospective observational study.

2.3. Setting and participants

Our series includes, between 2016 and 2019, patients who were evaluated for flank pain, identified to have B/L hydroureteronephrosis (HUN) on initial ultrasound (USG) of the kidney-ureter–bladder (KUB) region, subsequently contrast-enhanced computed tomography (CECT) KUB confirmation of PL, and underwent successful robotic-assisted bladder fat extirpation with B/L ureteric reimplantation with double J (DJ) stenting.

2.4. Preoperative assessment

Patients who presented with flank pain underwent initial USG KUB, which identified B/L HUN. The diagnosis was confirmed with CECT abdomen showing features suggestive of PL and B/L HUN secondary to distal ureteric compression (Fig. 1–3). Patients were evaluated with routine blood counts, renal and liver function tests, coagulation profile, electrocardiography, and chest x-ray. All patients had normal creatinine preoperatively. Uroflowmetry was performed in all patients; all had normal flow patterns and values.

Ethical committee clearance was obtained and informed consent was acquired from all patients.

2.5. Procedure

Initial cystoscopy was performed in all patients. Patients who had bullous changes in the bladder underwent transurethral resection biopsy to rule out adenocarcinoma and were subsequently taken up for robotic procedure after 2 wk. All patients underwent successful wide bladder fat extirpation (at the site of reimplantation) and B/L ureteric reimplantation (extravesical refluxing type) with DJ stenting.

2.6. Surgical technique

The patient was positioned in a low lithotomy position with steep Trendelenberg incline and 16Fr Foley catheter in situ. Both the legs of the patient were placed in Allen stirrups with antiembolic stockings and a sequential compression device. A pneumoperitoneum was created with a Veress needle. The standard W configuration of port placement for pelvic surgeries was done, and a DaVinci Xi robot, with all four arms and a 12-mm assistant port, was docked in all cases. Both ureters were identified at the level of iliac vessel

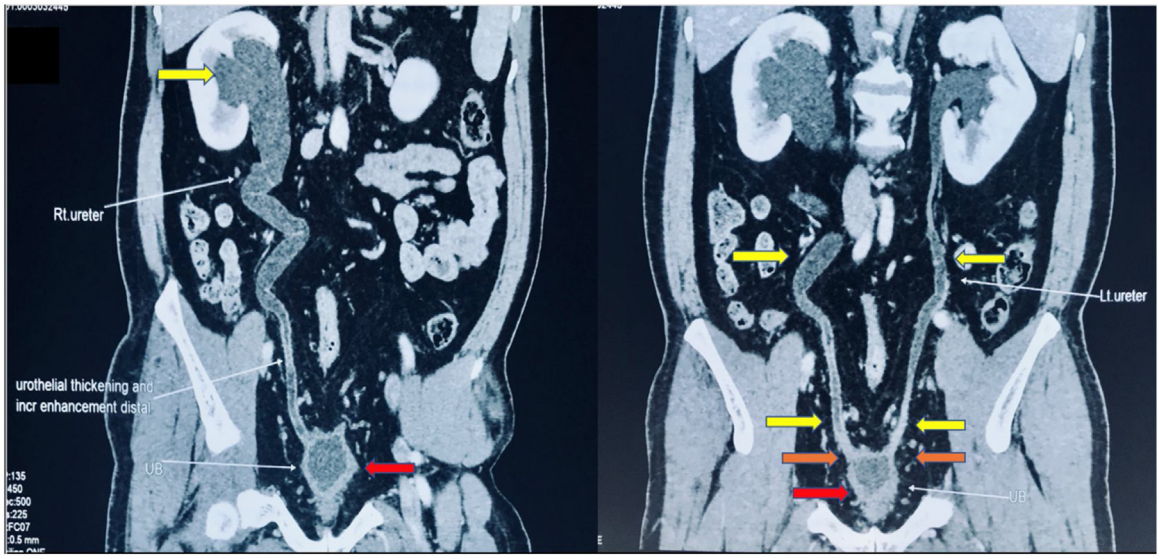

Fig. 1 – Bilateral distal ureteric thickening with segmental narrowing and bilateral hydroureteronephrosis (yellow arrows), straightening of bilateral vesicoureteric junction (orange arrows), and thickening of urinary bladder wall (red arrows).
Lt. = left; Rt. = right.

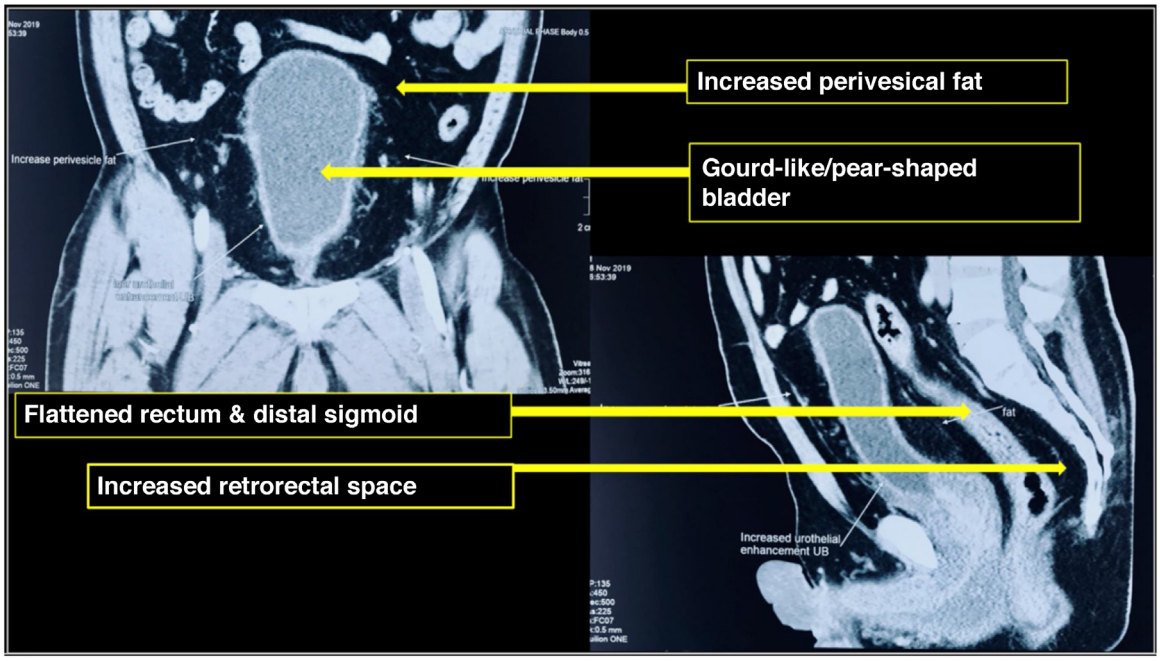

Fig. 2 – Typical CECT findings of pelvic lipomatosis demonstrated.
CECT = contrast-enhanced computed tomography.

crossing by opening the posterior peritoneum and carefully dissecting the fat layers. Ureters were dissected as distally as possible, clipped distally, and cut. The free ureteric margin was sent for histopathological examination (HPE). The bladder was distended with 200–250 cc saline, and ureteric reimplantation sites were marked on the dome of the bladder. Complete and wide extirpation of fat up to the bladder wall was performed at these marked sites, and the fat specimen was sent for HPE. The bladder wall was opened and B/L mucosa to mucosa ureteroneocystostomy was performed with 4-0 Vicryl over a 5Fr 26-cm DJ stent on either side (extravesical refluxing type). Bladder fat was closed loosely over the anastomosis and a 16Fr drain was placed (Fig. 4 and 5). Our recommended tips for the technique are described later in this article.

Complications were graded as per the revised Clavien-Dindo classification [8]. Postoperatively, patients were

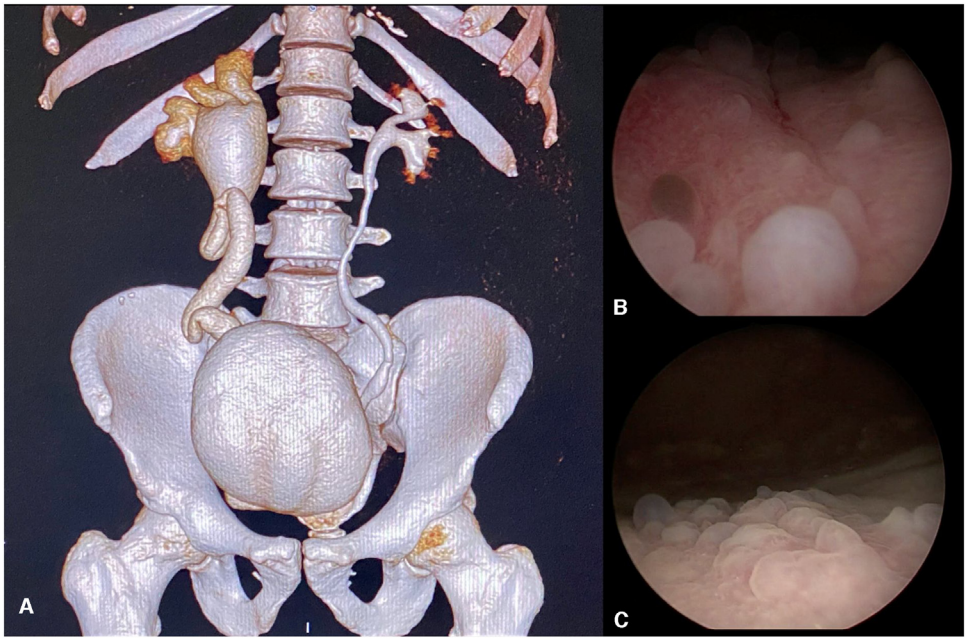

Fig. 3 – (A) Bladder out of pelvis corresponding to an elevated bladder neck on cystoscopy. (B and C) Bullous lesions seen at bladder neck and trigone region on cystoscopy.

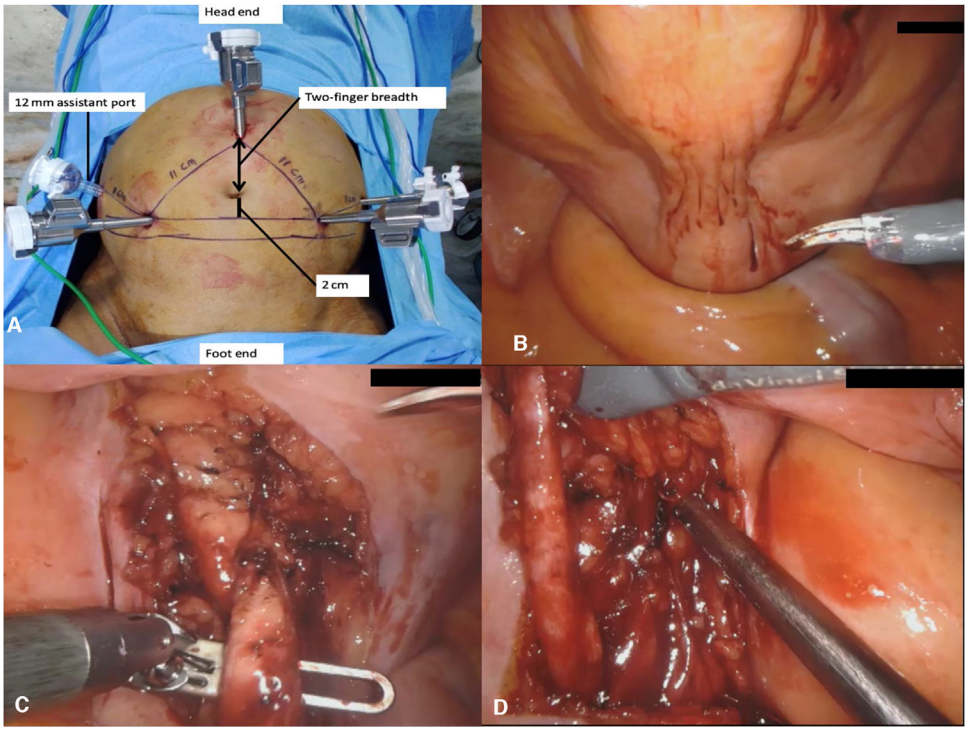

Fig. 4 – (A) W configuration of port placement. (B) Intraoperative appearance—the entire pelvis was filled with the bladder and fat. (C) Right and (D) left ureter dissection surrounded by thick and hypervascular fat, respectively.

ambulated and started on oral diet on postoperative day (POD) 1, and were discharged on POD 2 or 3 with the drain and the Foley catheter; the drain was removed on POD 5 or 6, and the Foley catheter on POD 10. All patients underwent B/L DJ stent removal at 3 mo.

2.7. Follow-up

All our patients are on regular follow-up with USG KUB, urine routine microscopy, and serum creatinine initially at 3 mo and annually thereafter till the end of the study period.

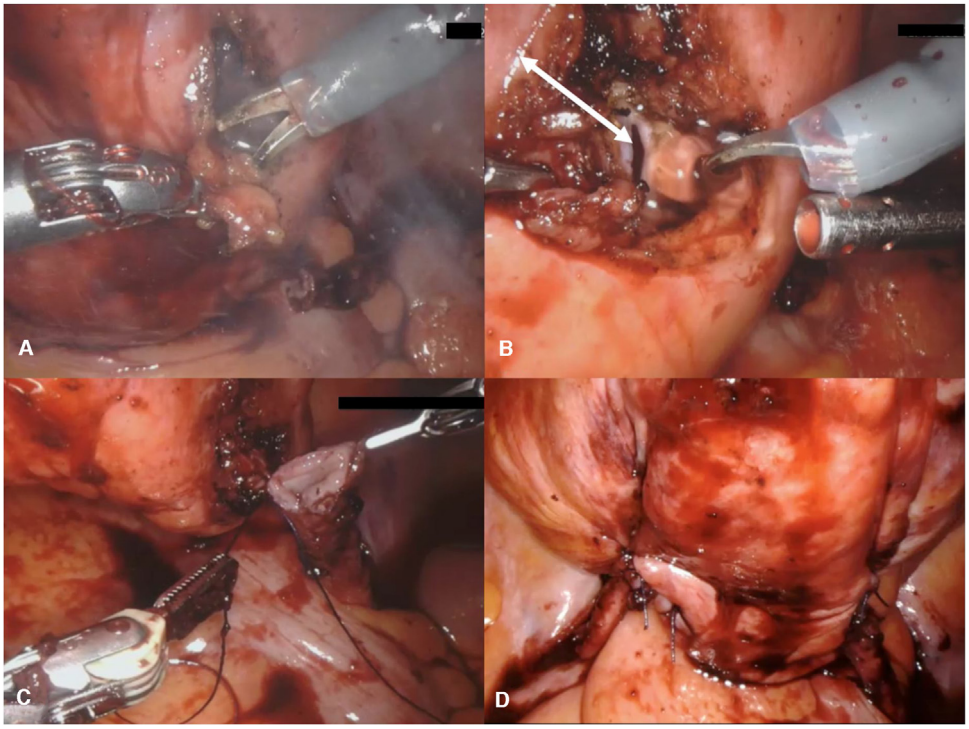

Fig. 5 – (A) Bladder fat extirpation at the site of ureteric reimplantation. (B) Bladder mucosa opened; thickness from surface to mucosa denoted by white arrow. (C) Mucosa to mucosa ureteroneocystostomy. (D) Final appearance after bilateral completed reimplantation.

2.8. Statistical analysis

Continuous variables are presented as median and range, as the sample size is very small.

3. Results

We encountered a total of five patients with PL during our study period. All our patients were male. The median age was 43 (range 36–56) yr and the median body mass index (BMI) was 26 (range 22–32) kg/m^2. All patients had normal serum creatinine within the hospital's reference range (0.8–1.3 mg/dl). CECT of the abdomen confirmed the diagnosis with usual radiological characteristics, except for one case, which was an intraoperative surprise where imaging did not have the typical features. Initial cystoscopy revealed elevated bladder neck in all patients, diffuse bullous cystitis changes in the bladder neck; trigone regions in two patients; and in addition, both ureteric orifices were unidentifiable in one patient (total three; Fig. 3). All these three patients underwent transurethral resection and biopsy. HPE was reported as proliferative cystitis with cystitis cystica in two patients and additionally with foci of cystitis glandularis in one patient. They were taken up for surgery after 2 wk. All the patients underwent robotic-assisted wide bladder fat extirpation and B/L ureteric reimplantation (extravesical refluxing type) with DJ stenting. The median console time was 126 (range 120–130) min, and the median blood loss was 120 (range 100–150) ml. Postoperative complications were Clavien-Dindo grade I in three cases, and the median hospital stay was 2 (range 2–3) d. Distal ureteric margins showed subepithelial edema with submucosal fibrosis, and bladder fat biopsies were reported as adipose tissue with hemorrhage. At initial 3-mo and annual follow-ups, all patients had normal serum creatinine and there was no evidence of disease recurrence (Tables 1 and 2).

Table 1 – Demographic data

Case	Age (yr)	BMI (kg/m^2)	Symptoms	Imaging identified	Type of surgery
1	56	25	FP with B/L HUN	Yes	Robotic
2	40	31	FP with B/L HUN	Yes	Robotic
3	36	32	FP with B/L HUN	Yes	Robotic
4	43	22	FP with B/L HUN	No	Robotic
5	48	26	FP with B/L HUN	Yes	Robotic

B/L HUN = bilateral hydroureteronephrosis; BMI = body mass index; FP = flank pain.

Table 2 – Results and observations

Sample	Parameters	Case				
		1	2	3	4	5
1	Preoperative creatinine (mg/dl)	0.9	1	1.1	0.8	0.9
2	Console time (min)	130	126	120	130	125
3	Blood loss (ml)	120	120	100	150	130
4	Hospital stay (d)	3	2	2	3	2
5	Drain removal (POD)	6	5	5	6	5
6	Foley removal (POD)	10	10	10	10	10
7	Postoperative complications (Clavien-Dindo grade)	I	I	–	I	–
8	Follow-up period till date (yr)	3	3	2	1	2
9	Follow-up creatinine at (mg/dl):					
	3 mo	1	1.1	1.1	1	1
	1 y	0.9	1	1	0.8	1
	2 yr	1	1	0.9	NA	0.9
	3 yr	1	0.8	NA	NA	NA
10	Outcome	NER	NER	NER	NER	NER

NA = not applicable; NER = no evidence of recurrence; POD = postoperative day.

4. Discussion

PL is considered a rare progressive benign disease. So far, only <150 cases have been reported worldwide, including only two single-center series (with 35 and eight cases) as per the available literature. A PUBMED and MEDLINE database search with keywords “pelvic lipomatosis” and “surgery” revealed a total of 51 articles only. Predominantly, it has been a disease of the males, with male to female ratio being 1.8:1 [9]. In our series, all the patients were male. The most commonly affected age group is between 20 and 60 yr, with only two pediatric cases reported so far [10,11]. Our patients’ age ranged between 36 and 56 yr, with a median age of 43 yr.

Although the etiology and natural course of PL remain unestablished, studies have quoted its relation to obesity. As per the World Health Organization guidelines for obesity for the Asia-Pacific region where individuals with a BMI of >25 kg/m^2 are considered obese, three of our patients were obese with a median BMI of 26 (range 22–32) kg/m^2 [12]. A possible genetic etiology, an abnormality in chromatin-regulating HMGA proteins, has been suggested. Overexpression of HMGA-2 and underexpression of HMGA-1 have been found to cause increased adipose tissue deposition in murine models [13,14].

Pain (flank or lower abdominal) is the most common symptom, and the wide range of other symptoms is secondary to the compression of pelvic structures—the genitourinary tract (lower urinary tract symptoms, hematuria, HUN, stones, and painful ejaculation), lower gastrointestinal tract (constipation, tenesmus, and rectal bleeding), and vascular system (edema of lower limbs and deep vein thrombosis) [4,15,16]. Complications include hypertension, renal failure, rarely bladder adenocarcinoma, pulmonary thromboembolism, portal vein thrombosis, and death [17–22]. All our patients presented with B/L flank pain (usually more on one side), and none had azotemia or hypertension. It has been reported that nearly 70% cases are associated with proliferative diseases of the bladder, such as cystitis glandularis, which is considered a precursor of adenocarcinoma of the bladder, emphasizing the importance of long-term regular follow-up [23–25]. In our series, proliferative cystitis with cystitis cystica was seen in two patients and additionally with foci of cystitis glandularis in one patient.

Historically, intravenous urogram has been used in the diagnosis of PL. With the evolution of technology, CT and magnetic resonance imaging have replaced intravenous urogram in the diagnosis, and USG is used more frequently in the initial phases of evaluation. Altered shape of the bladder (pear/vertically oval/banana shaped/gourd shaped) is a widely accepted distinctive feature of PL, with sensitivity and specificity of 40.6% and 100%, respectively [26]. Other features include increased perivesical and perirectal fat, flattened rectum and distal colon, bladder projecting out of the pelvis, and increased retrorectal space [27]. In addition to B/L HUN (secondary to distal ureteric compression), most of the features were demonstrable in our series except for one case, which was an intraoperative surprise (Fig. 1–3).

Multiple morbid procedures for urinary diversion, such as ileal conduit with or without simple cystectomy, cutaneous ureterostomy, and B/L percutaneous nephrostomies, have been performed in the past to treat B/L HUN. It is difficult for the young patients to accept such morbid procedures for a benign disease, considering their life expectancy and future quality of life. Open B/L ureteric reimplantation was offered increasingly, but no data are available on its long-term outcomes [8]. First, such a procedure was reported by Halachmi et al [28] in 1996 using an ultrasonic-assisted lipectomy device. Ge et al [7] reported promising results in a series of eight patients who underwent laparoscopic bladder fat extirpation and B/L ureteric reimplantation with a median follow-up of 48.5 mo, and only one patient had recurrence at 49 mo. However, long-term efficacy is not available. We have performed robotic-assisted wide bladder fat extirpation and B/L ureteric reimplantation with DJ stenting in five patients

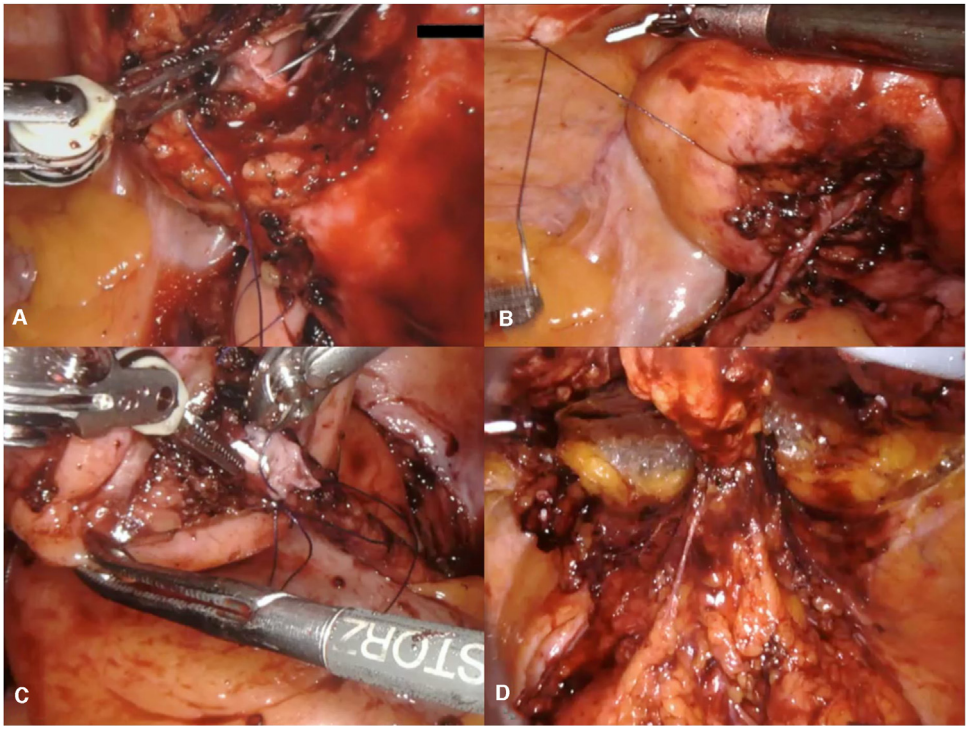

Fig. 6 – (A) Mucosal eversion stitch. (B) Tacking bladder wall fat to lateral pelvic wall. (C) Medial retraction of bladder wall fat by the assistant. (D) Bladder dropped from the abdominal wall, with bleeding from multiple points.

with promising short-term success. Although ureteric reimplantation has a high success rate in experienced hands, the major challenges encountered in surgery for PL includes the following: (1) lack of surgical planes, as the entire pelvis will be filled with fat, and difficulty in identifying the anatomic structures; (2) a large bladder occupying the entire pelvis, needing adjustment of port positioning and redocking; (3) presence of tough, thick, adherent, and hypervascular fat planes that bleed easily on manipulation; (4) difficulty in separating the ureter from adherent tough fat; and (5) bladder mucosa buried underneath the thick fat planes, making anastomosis difficult.

Apart from the well-established postulates of robotics compared with laparoscopy in pelvic surgeries, the specific advantages of performing this procedure with robotic assistance includes the following: (1) ease of operating in a narrow pelvic space in an ergonomic way, (2) camera completely controlled by the surgeon, (3) ease of mucosa to mucosa suturing in ureteroneocystostomy due to dexterity of wristed instruments, (4) ease of DJ stent placement, and (5) using the fourth arm for retracting the dropping bladder, ureter dissection as distally as possible to achieve an adequate-length tension-free anastomosis.

We recommend the following techniques, which we found helpful in successfully completing the surgery (Fig. 6):

1. Ureter identification at the level of iliac vessels, which can be identified with relative ease.
2. Complete and wide bladder fat extirpation at the site of intended ureteric implantation.
3. Performing mucosal eversion stitches when bladder mucosa is deeply buried underneath the thick fat planes.
4. Medial or lateral retraction of the surrounding fat screens of the bladder with sutures to lateral pelvic wall or by assistant help in adequate exposure for ureteroneocystostomy.
5. Wide ureteric lumen for anastomosis; usually the ureters are dilated with adequate luminal diameter for anastomosis, and when caliber is inadequate, oblique trimming of distal ends is recommended.
6. Ureter dissection as distally as possible with limited proximal mobilization, to retain its vascularity and to achieve tension free anastomosis.
7. Dropping the bladder from the abdominal wall when the anastomosis is in tension, but it could lead to heavy bleeding.

Since no specific guidelines are available for follow-up, we follow up our patients with USG KUB, urine routine microscopy, and serum creatinine initially at 3 mo and annually thereafter. All our patients have been doing well till the end of the study period, with no evidence of disease recurrence.

Limitations of this study are as follows:

1. The study has a very small sample size due the very low incidence of PL, although this is the third largest single-center series as per the available data.

2. Follow-up time is short and hence the question that how long the surgical effect will be sustained due to abnormal proliferation of fat cells remains unanswered; longer follow-up periods are needed.

5. Conclusions

PL causing B/L HUN has good surgical outcome. Imaging cannot always reliably detect this condition. We have had a good success rate and good outcome with this technique. However, the choice of surgical technique purely depends on the surgeon's comfort, leaving logistic issues aside.

***Author contributions*:** J. Sanjay Prakash had full access to all the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis.

Study concept and design: Sanjay Prakash, Jain.
Acquisition of data: Sanjay Prakash, Mathisekaran, Paul.
Analysis and interpretation of data: Sanjay Prakash, Paul, Selvaraj.
Drafting of the manuscript: Sanjay Prakash, Mathisekaran.
Critical revision of the manuscript for important intellectual content: Sanjay Prakash, Bafna.
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