ORIGINAL RESEARCH

Propensity-matched Analysis Comparing the Peri- and Post-operative Outcomes of Side-docking Versus Standard Lithotomy Docking for Robot-assisted Radical Prostatectomy

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Introduction: Limited access to the perineum and limited operating room space are just some of the limitations of the standard lithotomy docking for robot-assisted radical prostatectomy (RARP-LD). The side-docking technique (RARP-SD) may address these problems.
Methods: Thirty cases of robot-assisted radical prostatectomy were matched to 120 cases of RARP-LD cases by propensity scoring using age, body mass index (BMI), clinical T stage, biopsy Gleason score, and ultrasound prostate volume. Operative and docking time, complications were used to compare peri-operative and safety outcomes.
Results: Evaluation of 30 RARP-LD and 30 RARP-SD cases was done after propensity matching. Patient age, BMI, clinical T stage, biopsy Gleason score, and prostate volume were similar between the two groups ($p>0.050$). The mean docking time of RARP-SD is shorter than that of RARP-LD cases (7.56 vs. 4.12, $p<0.001$), but this did not translate to a shorter operative time. There were less peri-operative complications in the RARP-SD cases.
Conclusions: RARP-SD has a docking time and produces less complication than RARP-LD.

Key words: Robotic radical prostatectomy, prostate cancer, Side-docking

Introduction

Robot-assisted radical prostatectomy (RARP) may arguably be considered as the gold standard for surgical treatment of prostate cancer. Though it has been available for more than a decade, the technique of RARP has not ceased to evolve.[1] The robotic platform has enable urologists to develop new techniques of performing the surgery as well as how they use the robot.[2-5] While it is one of the less deliberated aspects of RARP, robot docking may play a major role in determining operative outcomes.

Historically, even while performing the open technique, the patient undergoing radical prostatectomy has been positioned in a Trendelenburg position with his legs lithotomy. However, the standard lithotomy docking for robot-assisted radical prostatectomy (RARP-LD) carries with it limitations that include restricted access to the perineum, a more challenging task of docking the robot, and its requirement for a larger operating room space.[6,7] The side-docking technique (RARP-SD) may address these problems.

Among the little that has been written about RARP-SD, most of the available literature has focused on describing docking time and neurologic complications related to positioning. To the best of our knowledge, no study has compared standard- and side-docking in terms of overall complications described using a standardized manner. In our institution, the rising concern for lithotomy-

related complications after RARP provoked the exploration of using side-docking for RARP cases. If proven to be associated with a decreased risk of position-related complications, side docking may prove to be a reasonable alternative to the standard lithotomy approach. In this article, the authors describe their initial experience with RARP-SD.

Methods

This was a retrospective analysis of prospectively-collected data of 125 cases done in a single tertiary institution from 2010 to 2017. After excluding the patients who had previous transurethral prostate or urethral surgery (4 patients) and one patient who underwent a Retzius-sparing approach to prostatectomy, a total of 150 patients were included in this study. RARP-SD was done in 30 cases while RARP-LD was done in 120 cases.

The authors compared the peri-operative and safety outcomes of RARP-LD and RARP-SD. Safety was measured by complication rates and blood loss. Peri-operative outcomes were measured by operative time, console time and docking time.

Standard docking was performed with the patient's legs abducted, partially flexed on stirrups and parking the robot placed in between the legs. During side docking, the legs were maintained straight slightly abducted and the robot was docked in the patient's right side, at a 45- degree angle to the patient's main axis. The patient was maintained in a Trendelenburg position in both docking techniques.

Docking time was measured from the time that the robot was rolled towards the patient from a parked position until all robotic arms were attached to the robotic trochars.

C-RARP was performed as previously described.[6] Posterior sphincter reconstruction was performed as described by Rocco.[7]

For the purpose of analysis, 1:1 propensity score matching was done between 30 RARP-SD cases and all RARP-LD cases using age, body mass index (BMI), pre-operative PSA, biopsy Gleason score (GS), and clinical T stage. Student's *t*- test and Pearson's x^2 test were used for analysis of quantitative and qualitative variables, respectively.

All statistical analyses were performed using the SPSS version 20 software (IBM Corp., NY, USA). A p value <0.050 was considered statistically significant for all two-sided tests. This study was approved by our Institutional Review Board.

Results

The pre-operative clinico-pathological characteristics of the two groups are presented in Table 1. No significant differences in terms of the variables used in the propensity score matching (patient age, BMI, pre-operative PSA, biopsy GS, and TRUS prostate volume) existed between the two groups.

Table 2 shows the peri-operative outcomes. Docking time was significantly shorter in the side docking group. Total operative time, console time, and blood loss were not significantly different for both groups. There were significantly more complications associated with RARP-LD. For the RARP-LD group, there were five Clavien-Dindo class II complications (1 case of deep venous

Table 1. Pre-operative characteristics.

	Standard docking N=30	Side docking N=30	p-value
Age (years) ± SD	62.93 ± 8.31	61.82 ± 8.56	0.826
BMI (kg/m2) ± SD			
Pre-operative PSA (ng/dl) ± SD	19.79 + 13.92	19.03 + 16.89	0.354
Biopsy Gleason score (%)			0.061
6	14 (46.7)	7 (23.3)	
7	9 (30.0)	17 (56.7)	
≥8	7 (23.3)	6 (20.0)	
Clinical stage (%)			0.767
T1	0	0	
T2	28 (93.3)	29 (96.7)	
T3	2 (6.7)	1 (3.3)	
Prostate volume (g) ± SD	44.86 + 28.85	43.68 + 16.29	0.443

SD= standard deviation, BMI = body mass index, PSA = prostate-specific antigen, * = significant p-value

Table 2. Peri-operative characteristics.

	Standard docking N=30	Side docking N=30	p-value
Total operative time (min) ± SD	369.33 + 88.04	354.06 + 74.26	0.905
Console time (min) ± SD	327.00 + 121.86	310.43 + 87.06	0.483
Docking time (min) ± SD	7.56 + 3.07	4.12 + 1.36	<0.001*
Estimated blood loss (ml) ± SD	672.33 + 482.99	677.78 + 532.75	0.948
Pathologic Gleason score (%)			0.109
6	14 (46.7)	10 (33.3)	
7	14 (46.7)	17 (56.7)	
≥8	2 (6.6)	3 (10.0)	
Pathologic stage (%)			0.767
T1	0	0	
T2	28 (93.3)	29 (96.7)	
T3	2 (6.7)	1 (3.3)	
Complications (%)			
All types	9 (23.3)	1 (3.3)	0.024*
Complications related to positioning	6 (20.0)	0	0.010*
Clavien-Dindo (%)			0.009*
I	3	1	
II	2	0	
III	3	0	
IV	1	0	
Hospital stay (days) ± SD	6.10 ± 2.78	4.82 ± 1.42	0.111

SD= standard deviation, * = significant p-value

thrombosis, 4 cases of neuropraxia). There was only one case of Rhabdomyolysis (class IV complication) with an incidence of 3.3% in the RARP-LD group and 1.7% overall. This rhabdomyolysis patient developed renal failure, underwent dialysis and was discharged 16 days after surgery. Complications not related to patient positioning included one case of post-operative bleeding necessitating cystoscopy with clot evacuation, one case of anastomotic leak, and one case pelvic abscess that required a pigtail insertion for drainage. The sole complication in the RARP-SD group was a case of post-op bleeding that necessitated blood transfusion.

Discussion

In this study, the authors compared the peri-operative, and safety outcomes between RARP-LD and RARP-SD. Several advantages of RARP-SD were observed in their study.

In their study, docking time was significantly shorter in the SD-RARP group. This finding is similar to the findings of previous studies.[8,9] This difference, however, did not translate to a significant difference in total operative time. Failure to observe differences in total operative time is due to the fact that docking time comprises a very small portion of the total operative time. In addition, total operative time is clearly dependent on several other factors such as difficulty of the surgery and surgeon experience. While significant differences in docking time may not have much effect on the total operative time, it is not without importance. Docking the robot can be one of the most complicated steps during RARP. In their study examining the association between intra-operative flow disruption and teamwork, Weigl, et al showed that the highest disruption in the flow of the operation occurred during the docking phase.[10] Shorter time of docking in RARP-SD can be taken as evidence supporting that it is easier for the surgeon to coordinate with his assistant when performing this technique. For those new to the robotic platform, this has a potential to hasten the learning curve of robot docking.

The overall complication rate in the present study is akin to those reported in available literature. Overall complication rate of RARP has been reported to be around 10%.[11,12] In their systematic review of 110 papers evaluating RARP outcomes, Novara, et al. noted that the most common complications were lymphocoele formation, urine leakage and reoperation. In contrast, complications in the present study population consisted mainly

of lower extremity peripheral neuropathy which is a Clavien-Dindo class I type of complication.

The most important result of the present study is the significantly lower complication rate of RARP-SD. Though still scarce, there have been studies that have shown less incidence of complications in RRAP-SD compared to RARP-LD.

Rhabdomyolysis is one of the most dreaded complications of prolonged Trendelenburg position. Patients who develop rhabdomyolysis are at increased risk of kidney injury and mortality, have prolonged hospital stays and spend more on treatment. In general, this complication has been found to occur rarely after RARP.[13,14] In their study of 60 patients who underwent RARP with extended pelvic node dissection, Mattei, et al reported an association between the Trendelenburg position and the occurrence of rhabdomyolysis.[15] Interestingly, the incidence of rhabdomyolysis that they reported (16.7%) is higher than what is here reported (1.3% overall) and those of other studies. One possible explanation their definition of rhabdomyolysis in terms of elevated post-operative creatinine values may have resulted in over-detection of the disease. Other than prolonged lithotomy position, other factors that have been linked to rhabdomyolysis are co-morbidities and BMI.[2]

Corneliu, et al. reported the incidence of peripheral neuropathies after RARP to be between 1.3 and 10.8%. They further illustrated that neuropathies were more commonly observed in the Lower. Comparable to the data in the present study, the incidence of peripheral neuropathy was noted to be at 6.7%.

Other reported advantages of docking the robot on the side of the patient include a better access to the perineum.[17] This is especially important in case a rectal injury does occur. Since the robot in on the patient's side, there is easier access to the perineum without needing to break sterility. In a more practical sense, side-docking saves a lot of space and is therefore more suitable for small operating theaters.[1]

In this study, SD-RARP was shown to have shorter docking time and less complications. While it is still too early to recommend that robotic prostatectomies be done using the side docking technique, it may prove useful to surgeons who wish to improve their peri-operative complication outcomes. It also has the potential to shorten the robotic team's learning curve for docking the robot.

This study is not without its limitations. First, this study still suffers from a modest sample size. Additional, better-powered studies are needed to support present findings. Secondly, results were based on consecutive cases performed by a multiple-surgeon cohort. Inter-observer bias as well surgeon's position in his learning curve may have affected the results. Randomized controlled trials comparing RARP-LD and RARP-SD should produce a more accurate analysis. However, the impetus in this institution to shift to the side-docking technique was on the authors' observation of an increasing number of complications that they attributed to patient positioning. Given the satisfactory results of RARP-SD so far, it would not be in their patient's best interest to revert back to the standard lithotomy approach just for the sake of randomization. Additional studies with greater analytical power that include functional outcomes and analysis of treatment cost for complications are still needed.

Conclusion

The advantages of RARP-SD, compared to RARP-LD include a faster docking time and less overall and position-related complications.

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ORIGINAL RESEARCH

Practice Variations for Surgical Oncological Cases Among Adult Urologists in the Philippines in the Management of Post-Surgical Reconstruction and Complications*

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Objectives: To identify practice variations among adult urologists in the surgical management of their oncologic cases and postoperative complications.
Methods: Beginning March 2022 to October 2022 an internet-based survey was performed among members of the PUA practicing in the Philippines.
Results: 82 Philippine urologists answered the survey during the study period. Majority have no subspecialty training (n=42) and practice primarily in the NCR (n=49). Open radical prostatectomy is the option of choice (n=58) with reported incidence of complications similar to that of previous studies. Conduit (n=77) is the diversion of choice after radical cystectomy with the majority recommending a two-surgeon approach in the harvest and reconstruction.
Conclusion: Practice is focused within the NCR with the majority having no subspecialty training thus preferring open surgical approach and two-surgeon team. Implantable devices are the preferred method in managing erectile dysfunction and urinary incontinence but is still lacking local availability

Key words: Urologic reconstruction, Uro-oncology, Philippine urology

Introduction

Prostate cancer is the third most common cancer and fourth leading cause of cancer deaths among males in the Philippines. It is the most common genito-urinary tract malignancy detected compared to bladder and testicular malignancies, each comprising one percent of new cancers detected.[1] Surgical management of these malignancies may include reconstruction and harvesting a segment of bowel, wherein other services may be needed. Post-operative complications are also varied depending on the surgery done, more so the management of these complications may need further surgical reconstruction or referrals to other services. No previous local report has been done regarding surgical approach for the management of these malignancies and its postoperative management. It was the goal of this study to determine these preferences.

Methods

An internet-based survey was conducted among adult urologists in the Philippines in March to October 2022. The questionnaire included a total

*This study was supported by the Philippine Society of Urologic Oncologists and the Philippine Society of Genitourinary Reconstructive Surgeons

of twenty questions. To participate in the survey the physician had to be a certified member of the Philippine Urological Association. All results were included in the analysis.

A population of 400 was determined based on the PUA master list of active practicing urologists in the Philippines. Sample size was then computed using the Cochrane formula with a confidence interval of 95% and a margin of error of 10%, yielding a sample size of 78.

Results

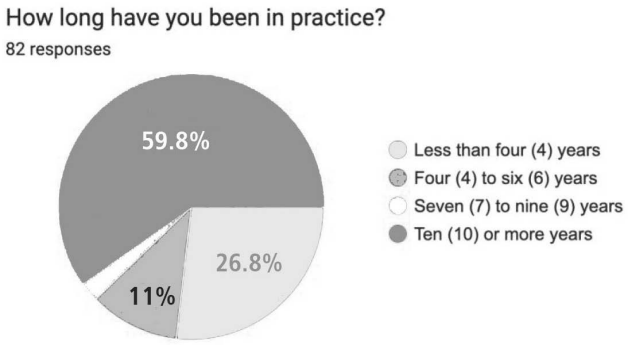
Demographics

A total of 82 responses were retrieved from the members of the Philippine Urological Association during the study period. Most respondents have been in practice for 10 years or more (n=49). This was followed by those who have recently started their practice for less than four years (n=22) (Figure 1). Most have not undergone subspecialty training (n=44) while others have multiple subspecialty training (n=19). The predominant subspecialty training is in the field of Oncology (n=14) followed by Endourology (n=12) and Laparoscopy & Minimally Invasive Surgery (n=11) (Figure 2).

Practice is focused within the National Capital Region (NCR) (n=49), wherein majority (35) practice solely in the NCR. Extension of practice from the NCR would reach Central Luzon (Region III) (n=7) and to Southern Tagalog (Region IV) (n=7). Other respondents reported practice would go as far as Ilocos (n=1) or Mindanao (n=1). Among the respondents who practice solely outside of NCR, the majority practice in Southern Tagalog (n=7). There was an equal distribution of respondents from Bicol (n=4), Central Luzon (n=4), and Central Visayas (n=4) (Figure 3). Almost all hold practice wherein there is either a General Surgery or Urology residency program.

Patient Case Load

Majority report of handling less than three prostate surgical cases per month (n=48). This was followed by four to six cases (n=24) with a few reporting seven to nine cases (n=2), and ten or more cases (n=8). The same trend holds true for bladder (n=70), penile (n=81), and testicular (n=76) oncologic cases but to a greater degree.



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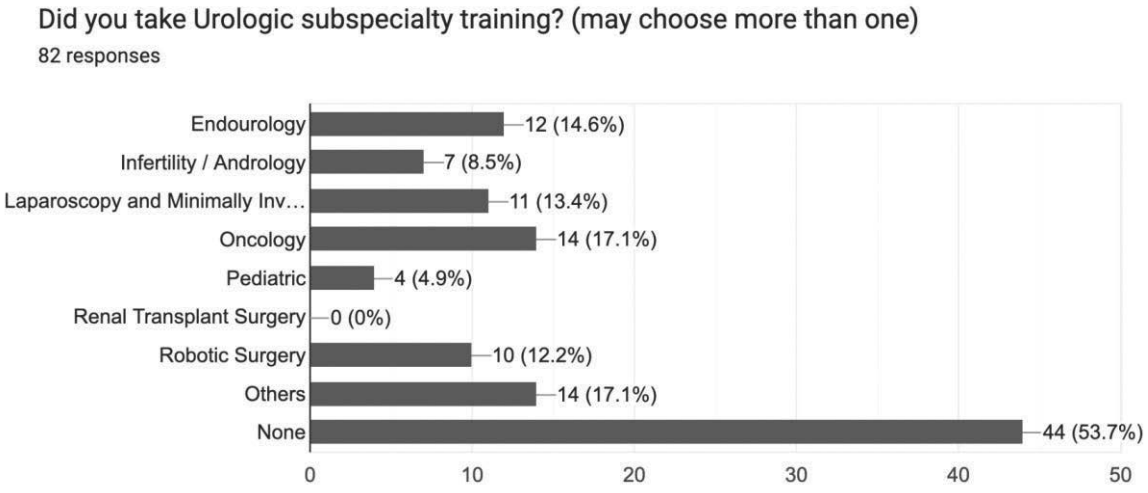

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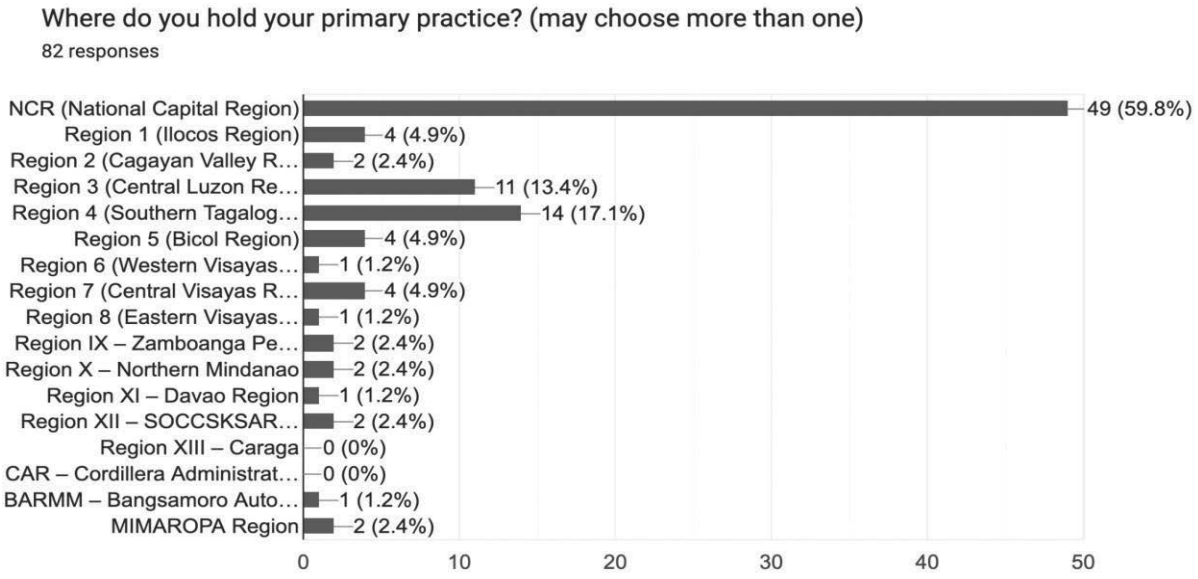


Figure 3.

Prostate Oncologic Practices

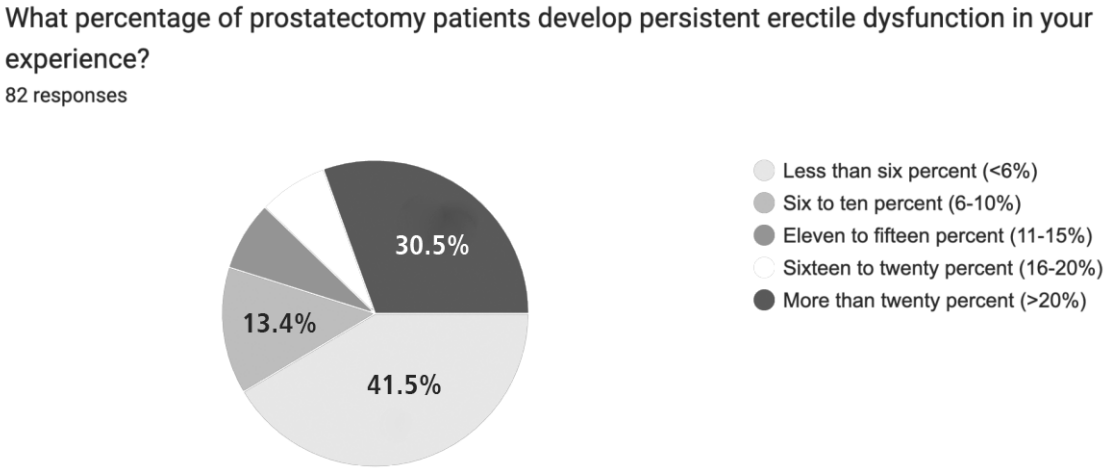
Open prostatectomy (n=58) is the preferred surgical approach in the Philippines. The remaining adopted laparoscopy (n=13) or a robot-assisted (n=9) approach. There were reports of laser resection (n=1) or referral to another surgeon (n=1) for the surgical management of prostate cancer.

Erectile Dysfunction After Prostatectomy

The reported incidence of erectile dysfunction (ED) after prostatectomy varied as most reported less than six percent (n=34) followed by those reporting more than twenty percent (n=25) (Figure 4). Almost half would recommend a semi rigid / malleable prosthesis (n=41) in the management of ED. Some would recommend two-component prosthesis (n=8) or three-component prosthesis (n=7). Other reported management options included medical management (n=12) or referrals to other urologists or institutions (n=4) (Figure 5).

Persistent Incontinence After Prostatectomy

Incontinence after prostatectomy has been reported to be as low as less than three percent (n=54) (Figure 6). Preferred management for persistent incontinence would be via artificial urinary sphincter (n=36). The remaining options



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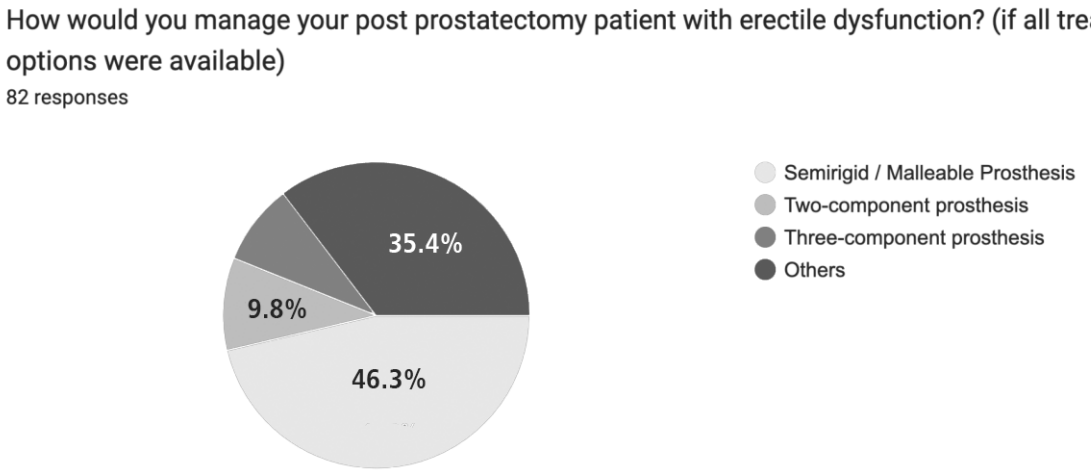

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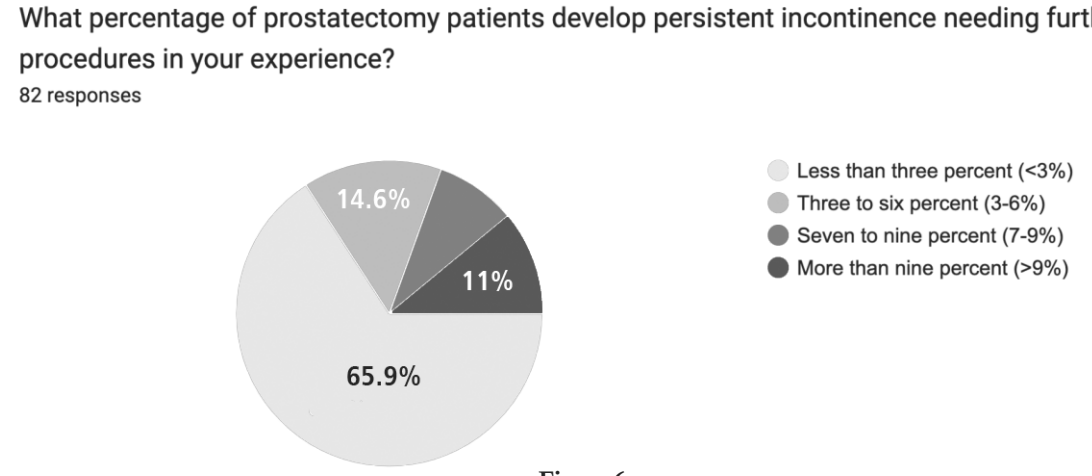


Figure 6.

are fairly even in preference: penile clamp (n=11), sling (n=8), and bulking agents (n=5). Others will continue conservative management (n=14) via continued medical management, Kegels training, or sanitary pads) (Figure 7).

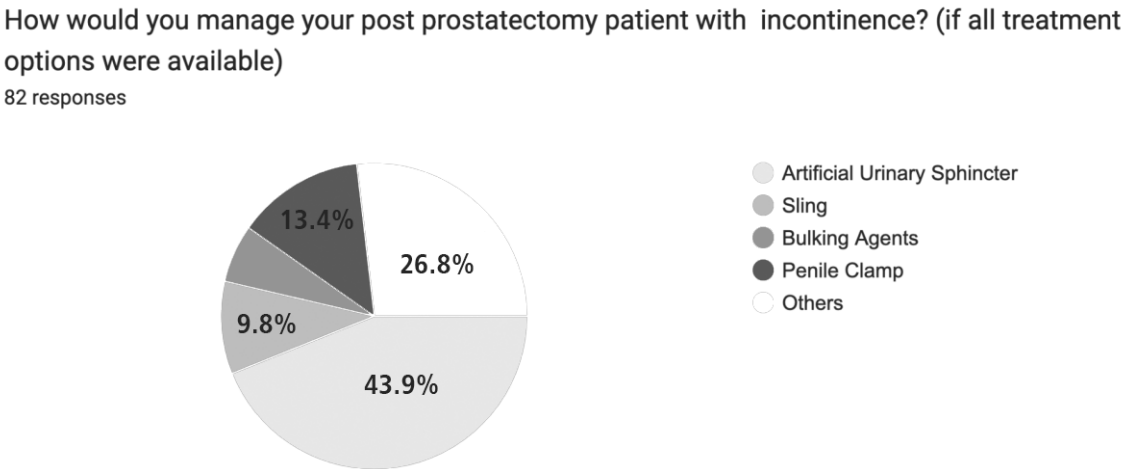
Bladder Oncologic Practices

Radical cystectomy would require creation of a new reservoir or diversion of the urine. Majority practice the creation of an ileal conduit (n=77). Only a few would consider an orthotopic neobladder (n=5) or a urinary reservoir (n=1) (Figure 8). During creation of the ileal conduit most prefer to do the harvest themselves alongside another urologist (n=39) or alongside a urologic resident (n=23). Referral to a general surgeon (n=17) or another urologist (n=3) are also options considered (Figure 9). Once the segment is harvested, the majority would prefer a hands-on approach in the reconstruction alongside another urologist (n=46) or with a urologic resident (n=31) (Figure 10).

Urinary diversion after radical cystectomy has its own set of complications. Metabolic complications are expected and most would employ a referral system to Internal Medicine or a Multidisciplinary approach (n=62). A fraction of the respondents prefers to independently (n=20) manage these metabolic complications (Figure 11).

Stricture After Surgery

Prostatectomy and orthotopic neobladder patients can possibly develop a urethral stricture



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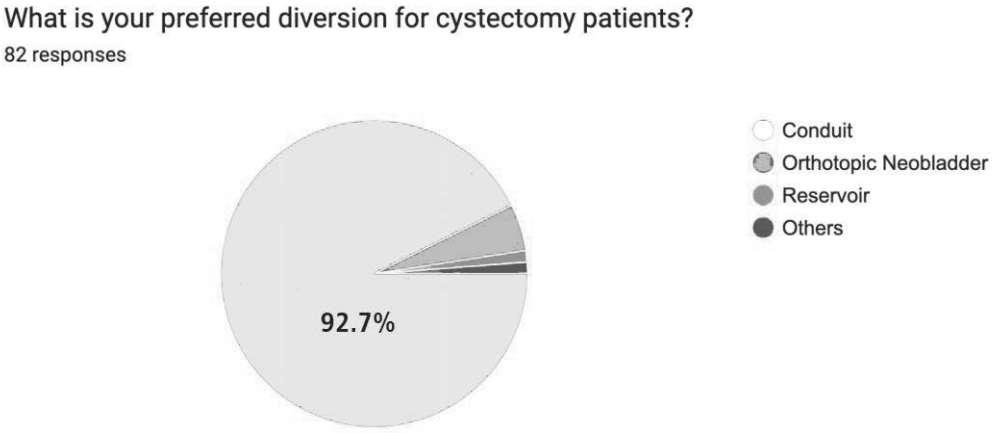

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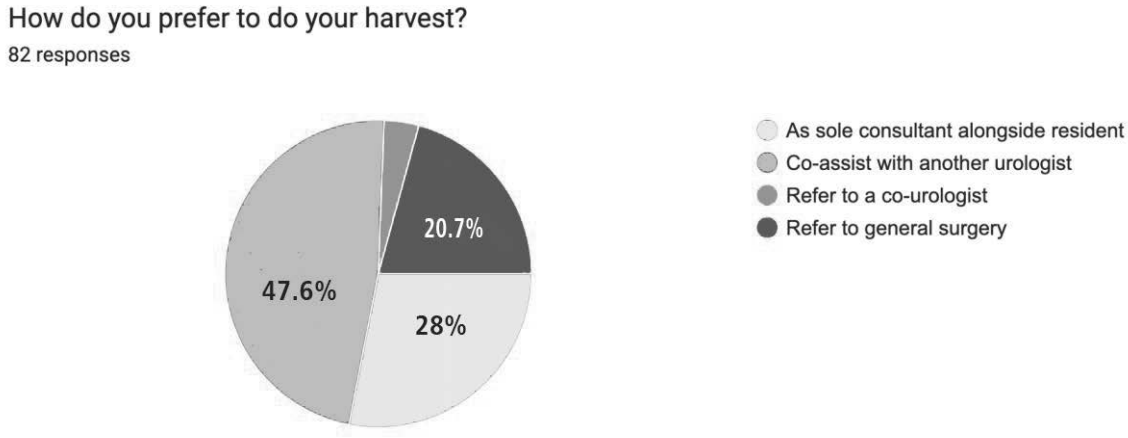

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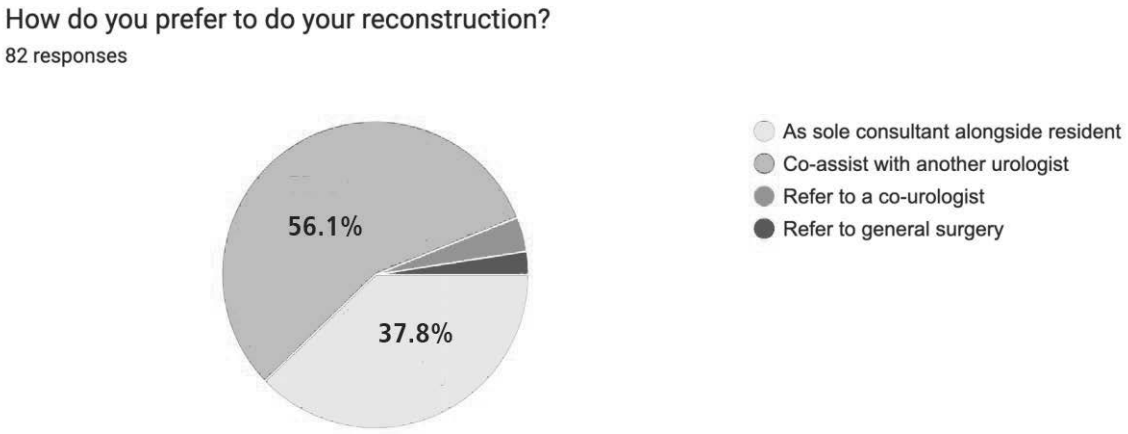

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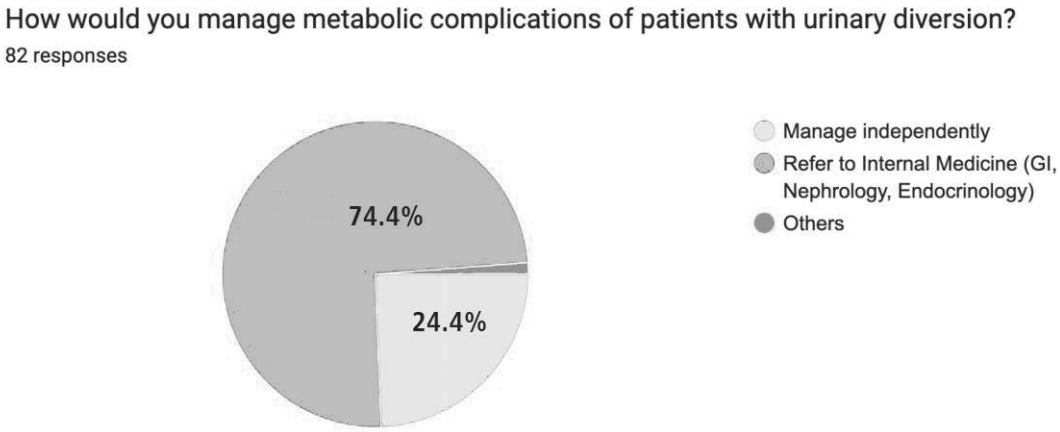

Figure 11

and/or a bladder neck contracture. Majority would manage this via endoscopy through direct visual internal urethrotomy (n=66) or transurethral incision of bladder neck (n=74). The remaining would recommend diversion (n=6) or open surgery with reconstruction / urethroplasty (n=5) in the management of urethral strictures (Figure 12), while buccal graft reconstruction (n=7) and open correction (n=2) of the anastomosis is an option for bladder neck contractures (Figure 13).

Penile Oncologic Practice

As the majority handle only a few penile oncologic cases, the majority would refer the reconstruction to either a co-urologist (n=29) or to a plastic surgeon (n=27). Those who do handle the reconstruction prefer the usage of a flap (n=14) or a graft (n=8). The remaining would stop after urethrostomy (n=2) (Figure 14).

Discussion

The data collected shows that the Philippines has a concentration of Urologists within the greater NCR and that most have no subspecialty training. This could be because that most respondents are those who have been in practice for a longer period of time, a time when subspecialty training was building up and those who have recently just started their practice wherein they have not yet been able to undergo further training.

More prostate oncologic cases are handled monthly by the respondents. This is in accordance with recent statistics reporting prostate cancer as ranking fifth in the Philippines for newly diagnosed cases, compared to bladder (18th) and penile (32nd) cancer.[1] Yet, surgical management of all these cases is minimal. This is probably due to the privatization of practice in the Philippines, possibly low detection rates within the country, and multiple non-surgical treatment options for urologic oncology.

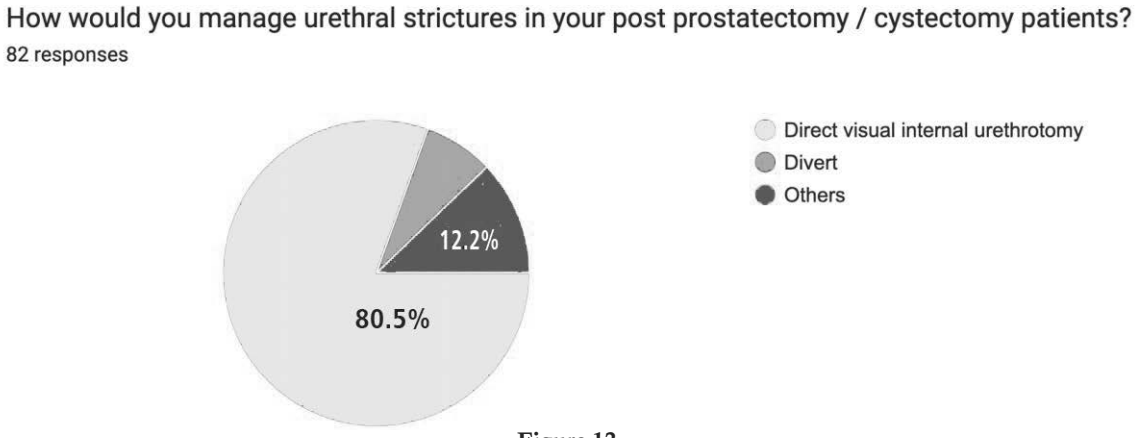

Figure 12

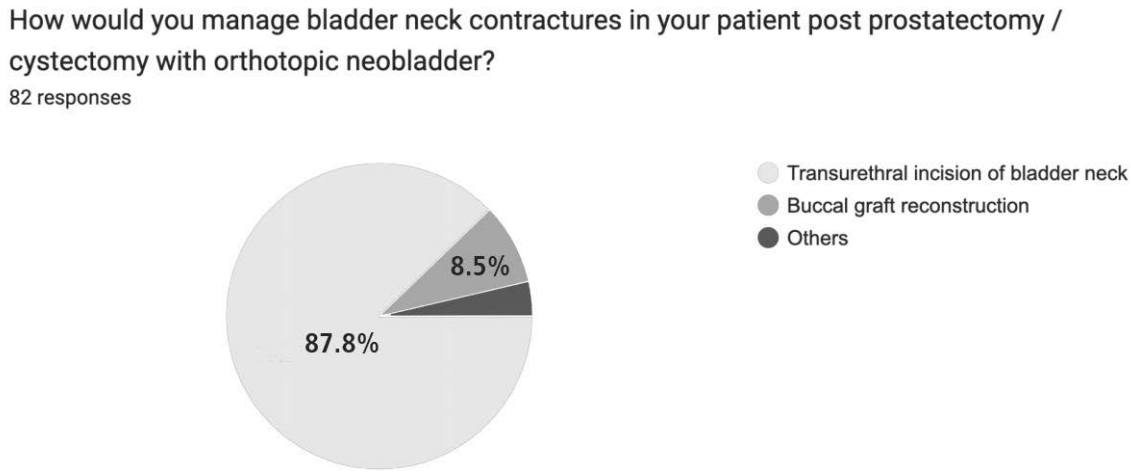

Figure 13

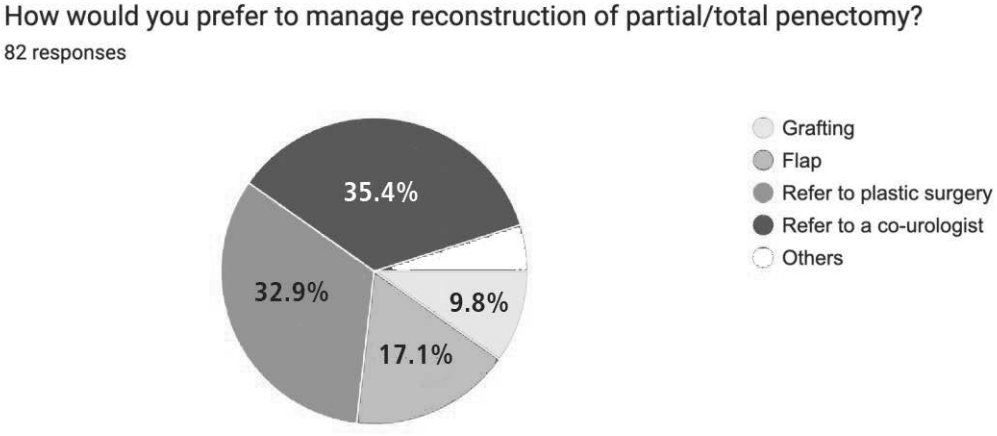

Figure 14

Radical prostatectomy (RP) is one of the treatment options for localized prostate cancer. The open surgical approach is still the favored approach in the Philippines. This could be that only a few respondents have undergone training in laparoscopy or robotic surgery.

One of the possible complications after RP is erectile dysfunction (ED). The survey showed a wide range of incidence in keeping with previous reports.[2] A multitude of treatment modalities are available for ED. These include phosphodiesterase 5 inhibitors, vacuum erection devices, intracorporeal injections, intraurethral therapy, and penile implants. This is initially managed conservatively as ED due to neuropraxia recovers over time, however for those with severe or persistent ED, penile implants reportedly have the highest effectivity and patient satisfactio.[2] Only a low percentage would eventually undergo implant after RP.[2] Factors that increase the likelihood of undergoing a penile prosthesis implant include lower age at diagnosis of prostate cancer.[3] There are two available prostheses in the market, the semirigid/malleable and the inflatable prosthesis, but only the former was previously given approval from the Philippine Food and Drug Administration.

A temporary urge incontinence is expected after RP due to neuropraxia, which more often than not recovers within the first year after surgery. Incidence of persistent urinary incontinence beyond this is low. Persistent incontinence varies depending on several factors such as pre-operative continence status, detrusor function, surgical factors.[4] Artificial urinary sphincter is still the gold standard in the management of persistent incontinence after RP[5], as such this is the recommendation of most. followed by continued conservative management via Kegel's exercises in conjunction with the use of sanitary pads or diapers.

Radical cystectomy entails the creation of a new urinary reservoir or diversion. The creation of an ileal conduit is the preferred choice among the respondents. Majority would still prefer a hands-on approach in the harvest and creation of the diversion, but would rather have another urologist assist in the procedure, despite the availability of a urology and/or general surgery resident within their institutions. A previous study was done by Ludwig, et al[6] that showed that a two-surgeon team had a shorter operation time, and lower anesthesia and operating room costs offsetted by the surgeon charges with no difference in perioperative complications compared to that of a single surgeon. Ileal conduit has its own set of complications[7] majority of which are metabolic. Some urologists would still manage these complications, however, for the most part would refer to a colleague in internal medicine to oversee the management of these metabolic complications.

Conclusion

The Philippines has a growing number of urologists but majority focus their practice in the NCR and have no further subspecialty training. It is recommended that practice is extended further out into other regions and subspecialty training pursued. This is seen since the majority still prefer the open surgical approach compared to laparoscopic or robot-assisted approaches. Persistent erectile dysfunction is most effectively managed with an implantable prosthesis but as of date, there is no available prosthesis in the market in the Philippines, which would support pursuance of approval and use. Lastly, during radical cystectomy, a two-surgeon approach is preferred despite availability of residents. This could be that this is private practice outside of their training institutions. Thus, it is again recommended that practice be extended outside NCR to be able to help with the preference of a two-surgeon approach for these cases.

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ORIGINAL RESEARCH

Analysis of the Clinical Efficacy and Safety of Percutaneous Nephrolithotomy in Patients with Anatomical Variations: A Single Center Retrospective Study

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Introduction and Objective: Percutaneous Nephrolithotomy (PCNL) is the standard of care for renal stones >2cm. Kidneys with anatomical disparities resulting from fusion (horseshoe), malrotation, ectopic location (allografts) and bifid collecting systems present as a challenge because variations in vasculature, calyceal rotation and intervening viscera may make percutaneous access treacherous. Reported here is the authors' experience with PCNL in these types of kidneys.

Methods: A chart review was done on all patients who underwent PCNL at the National Kidney and Transplant Institute (NKTI) from 2012-2016. Those with anatomical variations were identified and analyzed. Patient demographics (age, gender, co-morbidity) and stone characteristics (Guy's stone score, laterality) were summarized. Intraoperative parameters such as location of puncture site (upper, mid, inferior calyces), number of tracts (single vs. multiple), operative time, estimated blood loss (EBL), and length of hospital stay (LOS) were analyzed. The primary endpoints were stone-free and complication rates according to the Clavien-Dindo (CD) classification.

Results: A total of 1,657 PCNLs were performed during the study period, of which 42 had anatomical variants. The mean age was 45.2±8.8 (R= 28-65) with a male to female ratio of 3:1. There were 18 horseshoe (42.9%), 15 bifid (35.7%), 7 malrotated (16.7%) and 2 renal allografts (4.8%.); Laterality-wise, 28 (67%) were left-sided, 12 (29%) were right-sided and 2 (5%) had right-sided pelvic kidneys (allografts). The Guy stone scores were 3 and 4 in 13 (30%) and 29 (70%) patients, respectively. The mean stone diameter was 3.8±0.6 cms. (R=2.5-5.5). Majority, n=37 (88%) were treated with an upper pole access. Thirty-six (86%) needed a single tract and while six (14%) required multiple tracts (bifid pelvis). The mean operative time was 111.5±28.1 mins. (R=65-188), EBL was 461±278.4 cc (R=200-1300). LOS was 3.6±0.94 days (R=2-7). The stone-free rate was 95%. Twenty-five (59.5%) complications were documented. Fifteen (35.7%) had fever: Grade I CD, and 10 (23.8%) required transfusion: Grade II CD. There was no mortality.

Conclusion: PCNL still persists as the treatment of choice for nephrolithiasis in kidneys with variations in anatomy or position. A high stone clearance rate can be achieved while minimizing complications.

Key words: Percutaneous Nephrolithotomy (PCNL), anatomical variants, stone-free rate, complications

Introduction

Renal stone disease affects 10-20 % of the population worldwide. Previously, the surgical options for the treatment of renal calculi were limited to open stone surgery. However currently, different modalitites are now available such as extracorporeal shockwave lithotripsy,

retrograde intrarenal surgery and percutaneous nephrolithotripsy.

Percutaneous nephrolithotomy is a minimally invasive endoscopic procedure for removal of large renal stones via a nephroscope passing into the collecting system. It is now considered as the gold standard for renal calculi more than 2 centimeters in size.

Different renal anatomic variants can be diagnosed during adulthood, including ectopic or fused kidneys (e.g. horseshoe anomaly). In these kidneys, the incidence of stone formation is higher due to associated urinary stasis and infection. The application of PCNL to these kidneys (kidneys not located in their usual anatomical location or those with aberrant anatomical variants) can be very challenging due to their unusual location (ectopic) or aberrant position. Kidneys with anatomic variants have unusually positioned renal pelvis and calyces, and aberrant blood supply. Ureteropelvic junction obstruction (UPJO) may also be present resulting from dense fibrous tissue at the proximal ureter or a high-inserting ureter.

Unique challenges in PCNL may result from intervening visceral organs such as the small or large bowel, making them prone to injury during the initial access. Consequently, this may affect the choice of calyceal entry and the ability to clear the stones effectively. For this reason, many prefer to treat these anomalous kidneys with stones using conventional open surgery.

The suggested predisposing factors include the abnormal position of the renal pelvis and calyces, anomalous vasculature, distortion of the upper ureter or the ureteropelvic junction (UPJ) by a dense amount of fibrous tissue, and a high-inserting ureter with abnormally-positioned UPJ.

The objective of this study was to evaluate the clinical safety and efficacy of PCNL for patients with congenital renal anomalies performed in NKTI as well as to compare the demographic data and preoperative profile of patients with renal anatomical variants

Reported here is the authors' experience with percutaneous nephrolithotomy with kidneys having anatomic variations performed at a single center.

Methods

Study Design

This is a retrospective cohort study that evaluated the clinical safety and efficacy of PCNL in patients with renal anatomic variants.

Sample Size

Sample size was computed using prevalence of 0.05 and precision 0.10 with a result of 34. Sample size was computed using pROC package of R ver3.6.3

```
> power.roc.test(auc=0.960, sig.level=0.05, power=0.99)

     One ROC curve power calculation

         ncases = 6.178686
       ncontrols = 6.178686
             auc = 0.96
       sig.level = 0.05
           power = 0.99
```

Study Population

Records of patients who underwent percutaneous nephroilithotomy from 2012-2016 were reviewed. The sample size was computed based on the aforementioned formula.

Those with anatomical variations were identified and analyzed. Patient demographics (age, gender, co-morbidity) and stone characteristics (Guy's stone score, laterality) were summarized. Intraoperative parameters such as location of puncture site (upper, mid, inferior calyces), number of tracts (single vs. multiple), operative time, estimated blood loss (EBL) and length of hospital stay (LOS) were analyzed. The primary endpoints were stone-free and complication rates according to the Clavien-Dindo (CD) classification

Ethical Consideration

This study was conducted in accordance with ICH GCP guidelines and regulations and approved by the NKTI REC with approval number NKTI REC 2017-117

1. Informed consent

This study was limited to chart review and there was no interaction with the participant therefore informed consent was not obtained.

2. Confidentiality

Patients were assigned case numbers to ensure anonymity. Only the authors had access to any data obtained in the study. In the event of any publication, all information collected from the charts will be kept confidential.

Statistical Analysis

Counts and percentages were used to summarize the data in categorical form, while means and its standard error (SEM) for data in quantitative form. Fisher's exact test was used to associate the anomaly with gender, comorbidity, laterality, Guy's classification of stone, tract, and Modified Calvien score. Analysis of variance (ANOVA) was used to compare the duration of surgery and hospital stay according to anomaly.

P-values less than 0.05 indicate significant differences. All statistical tests were performed in R.

Results

A total of 1,657 PCNLs were performed during the study period, of which 42 had anatomical variants. The mean age was 45.2±8.8 (R= 28-65) with a male to female ratio of 3:1. There were 18 horseshoe (42.9%), 15 bifid (35.7%), 7 malrotated (16.7%) and 2 renal allografts (4.8%.); Laterality wise, 28 (67%) were left-sided, 12 (29%) were right-sided and 2 (5%) had right-sided pelvic kidneys (allografts). The Guy Stone scores were 3 and 4 in 13 (30%) and 29 (70%) patients, respectively. (Table 1)

The mean stone diameter was 3.8±0.6 cms. (R=2.5-5.5). Majority, n=37 (88%) were treated with an upper pole access. Thirty-six (86%) needed a single tract and while six (14%) required multiple tracts (bifid pelvis). (Table 2)

The mean operative time was 111.5±28.1 mins. (R=65-188), EBL was 461±278.4 cc (R=200-1300). LOS was 3.6±0.94 days (R=2-7). The stone-free rate was 95% and was assessed using neephroscopy and post PCNL fluoroscopy. Twenty-five (59.5%) complications were documented. Fifteen (35.7%) had fever: Grade I CD, and 10 (23.8%) required transfusion: Grade II CD. There was no mortality. (Table 3)

Table 1. Demographics of patients with renal anatomical variations who underwent PCNL.

Total	Anomaly				
	Allograft Kidney	Horseshoe	Malrotated Kidney	Bifid pelvis	
Number of Patients	42	2	18	7	15	p-value
Gender:						
Male	28 (66.7%)	2 (100%)	12 (66.7%)	3 (42.9%)	11 (73.3%)	0.460
Female	14 (33.3%)	0 (0%)	6 (33.3%)	4 (57.1%)	4 (26.7%)	
Comorbidity:						
Diabetes Mellitus	10 (23.8%)	2 (100%)	5 (27.8%)	2 (28.6%)	1 (6.7%)	0.180
Hypertension	6 (14.3%)	0 (0%)	3 (16.7%)	1 (14.3%)	2 (13.3%)	
Laterality:						
Right	15 (35.7%)	0 (0%)	6 (33.3%)	4 (57.1%)	5 (33.3%)	0.570
Left	25 (59.5%)	0 (0%)	12 (66.7%)	3 (42.9%)	10 (66.7%)	
Guy's classification of stone:						
(4)	31 (73.8%)	0 (0%)	17 (94.4%)	4 (57.1%)	10 (66.7%)	0.007
(3)	11 (26.2%)	2 (100%)	1 (5.6%)	3 (42.9%)	5 (33.3%)	

Table 2. Intra-operative factors of patients with renal anatomical variations who underwent PCNL.

		Total	Anomaly			
			Allograft Kidney	Horseshoe	Malrotated Kidney	Bifid Pelvis
Access:	Upper	37 (88.1%)	2 (100%)	18 (100%)	5 (71.4%)	12 (80%)
	Mid	4 (9.5%)	0 (0%)	0 (0%)	1 (14.3%)	3 (20%)
	Inferior	3 (7.1%)	0 (0%)	0 (0%)	1 (14.3%)	2 (13.3%)
Tract:	Single	36 (85.7%)	2 (100%)	18 (100%)	7 (100%)	9 (60%)
	Multiple	6 (14.3%)	0 (0%)	0 (0%)	0 (0%)	6 (40%)

Table 3. Outcome of PCNL on patient with renal anatomical variations.

	Total	Anomaly				p-value
		Allograft Kidney	Horseshoe	Malrotated Kidney	Bifid Pelvis	
Duration of Surgery (min)	111.5 ± 28.2	95.0 ± 10.0	114.0 ± 4.8	107.1 ± 8.8	112.7 ± 10.1	0.808
Duration of Hospital Stay (days)	3.6 ± 1.2	4.0 ± 0.0	3.8 ± 0.3	3.0 ± 0.4	3.5 ± 0.4	0.520
Modifed Calvien Score: 2 (Requiring Blood Transfusion)	10 (23.8%)	0 (0%)	5 (27.8%)	1 (14.3%)	4 (26.7%)	0.940

Discussion

In this study, the incidence of patients with anatomical variations who underwent PCNL is 0.02% and majority had Horseshoe Kidneys (43%). PCNL is still the considered standard of care for renal anatomical variations with kidney stones more than 2 cms and the upper pole access is still the preferred access especially in patients with a horseshoe-kidney or pelvic-kidney, owing to the inferior displacement away from the pleura and majority (88%) used the upper pole access.

Percutaneous nephrolithotomy (PCNL) is considered as treatment of choice for renal stones, and some upper ureteric stones. It has been performed since 1980s, with overall success rates exceeding 90%.[1] Improvements in technique and instruments have diminished complication rates associated with this procedure.[1] PCNL is technically very challenging in anomalous kidneys because the abnormal pelvicaliceal system results in difficulty in access. An abnormal relationship to the surrounding structures increases the incidence of visceral and vascular injuries. Fusion and malrotation anomalies are the most common types of renal abnormalities presenting with stones in clinical practice. The horseshoe kidney is the most common renal fusion anomaly.

Extracorporeal Shock Wave Lithotripsy (ESWL) and PCNL are the 2 most commonly used modalities for managing the stones in horseshoe kidneys. Ureteroscopy is used less often because of the technical challenges encountered with the altered renal anatomy. ESWL is the preferred modality for stones 2 cm in anomalous kidneys because the stone-free rate has varied from 72% to 92% in different series, and PCNL remains the reference standard for large stone burdens and ESWL-resistant stones.[2] The upper pole access is relatively safe in patients with a horseshoe-kidney

or pelvic-kidney, owing to the inferior displacement away from the pleura.[3] Osther et al. reported that access-failure for PNL was significantly more in patients with renal anomalies (5 %) when compared to normal kidneys (1.7%).[4] Mosavi-Bahar reported mild pleural injury in two patients[5], while Gupta et al.[2], and Ozden et al.[6], reported intercostal tube drainage in one patient. Similarly, pneumothorax was reported by Raj et al. in 6 %.[7] On the other hand, Shokeir et al. and Viola et al.[8] reported no pleural complications in patients with upper pole PCN in a horseshoe-kidney.

In a study done by Gupta et al (2009), all patients with renal congenital anomaly had complete clearane, , 89% via single tract and 11% via multiple tracts, with a mean operating time of 82 minutes and hospital stay of 3.2 days. In a study done by Osther, outcomes of PCNL in normal vs with renal anomalies were similar in terms of frequency of common complications but noted longer duration of surgery and multiple access tracts in renal malformation. They also noted longer duration of surgery for horseshoe kidneys compared to other renal anomalies, and longer hospital stay for ectopic kidneys.[9]

Osther et al. reported that access-failure for PNL was significantly more in patients with renal anomalies (5 %) when compared to normal kidneys (1.7 %) and according to Mosavi-Bahar, mild pleural injury was reported in PCNL in horseshoe kidneys while Gupta et al.[2], and Ozden et al.[6], reported intercostal tube drainage in one patient who had horseshoe kidney who underwent PCNL Similarly, pneumothorax was reported by Raj et al. in 6%. Due to this, different centers in the country often times opt to do the open approach for renal stones due to limited experience in doing endoscopic procedures in such anatomic variants. In the present study, all patients were treated with PCNL and no major complications were noted and ten out of the forty required blood transfusion.

Conclusion

Nephrolithiases in kidneys with variations in anatomy and position are challenging. The urologist should be prepared to apply minimally invasive techniques to remedy this. The present study provides additional support that PCNL still persists as the treatment of choice for nephrolithiasis in patients with this clinical condition. A high stone clearance rate can be achieved effectively while minimizing complications.

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CASE REPORT

The Use of Minimally-invasive Cortical Sparing Adrenalectomy as an Approach to Bilateral Adrenal Masses in a Patient with von Hippel Lindau Syndrome: Learnings from a Lower Middle-income Country Setting

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von Hippel Lindau syndrome is a rare genetic disease which may present with bilateral adrenal masses requiring surgical intervention. Previous practice at UP-PGH was to perform outright total adrenalectomy on pathologic adrenal glands and rely on lifelong steroid replacement for patients who had both adrenals removed. Presented here is a case of a patient diagnosed with von Hippel Lindau syndrome with bilateral adrenal masses, surgically managed initially with open adrenalectomy on the right side, followed by the first ever performed minimally invasive cortical sparing adrenalectomy at UP-PGH on the left side.

Key words: Adrenalectomy, von-Hippel Lindau syndrome

Background

von Hippel Lindau (vHL) syndrome is a rare inherited disorder involving a genetic mutation in the vHL gene. It is characterized by tumor formation in different parts of the body such as the eyes, brain, spinal cord, kidneys, pancreas, and adrenal glands. Adrenal tumors of patients with vHL are usually of a pheochromocytoma etiology.[1] Surgical excision of the pheochromocytoma remains to be the primary treatment for these sets of patients. Previous practice involved total adrenalectomy on pathologic adrenal glands. Difficulties arise in patients who require bilateral adrenalectomy as these patients would require a lifetime of steroid replacement. Steroid replacement requires close follow-up to monitor and adjust doses of exogenous steroids. Failure to do so may result in complications such as weight gain, loss of libido, overall depreciation of quality of life and the most dreaded and potentially fatal adrenal crisis.[2]

Cortical sparing adrenalectomy involves excision of the tumor and leaving normal-functioning adrenal tissue behind. Most important benefit of this procedure is that if enough adrenal tissue is left behind, patients may be spared from lifelong steroid therapy and avoid its possible dreaded complications.[3]

The Case

A 19-year-old male presented with hypertensive spikes and headaches 3 years prior to consult. He was initially managed with 3 anti-hypertensive drugs but was advised to seek consult at a specialty institution. Further workup included abdominal imaging which revealed bilateral adrenal masses, elevated 24-

hour urine metanephrine (3.782 mg/24 hours) and genetic testing which yielded a positive result for a pathologic gene, confirming vHL syndrome. Considering the elevated 24-hour urine metanephrine result and a confirmed diagnosis of vHL syndrome, our primary impression for the adrenal masses was pheochromocytoma.

Representative cuts of the abdominal CT scan are shown below (Figures 1 & 2). The right adrenal gland contained 2 tumors: a 4.3cm x 4.2cm mass on the lateral limb (plain HU: 25.2, absolute washout 53%, relative washout: 44%), and a 2.1cm x 2.1cm mass in the medial limb (plain HU: 27.1, absolute washout 70%, relative washout: 59%). On the contralateral side, the left adrenal gland contained a solitary tumor measuring 2.2cm x 2.0cm in its body (plain HU: 20.5, absolute washout 57%, relative washout: 57%)

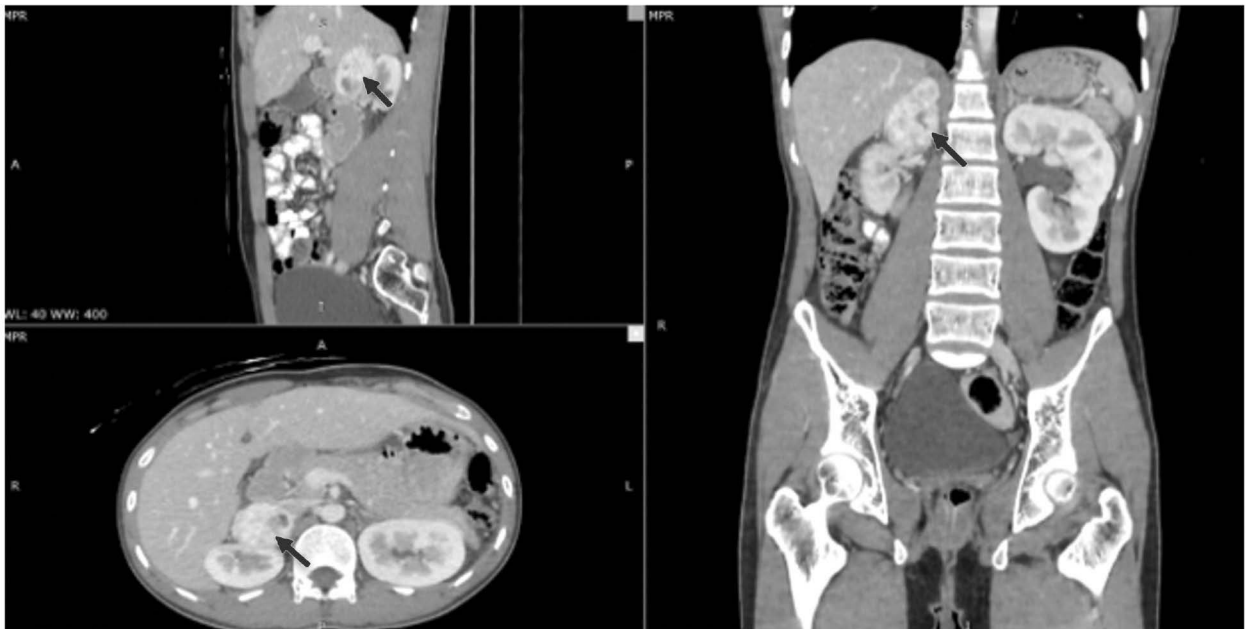

Figure 1. Pathologic right adrenal gland shown on different cuts from abdominal CT scan (arrows).

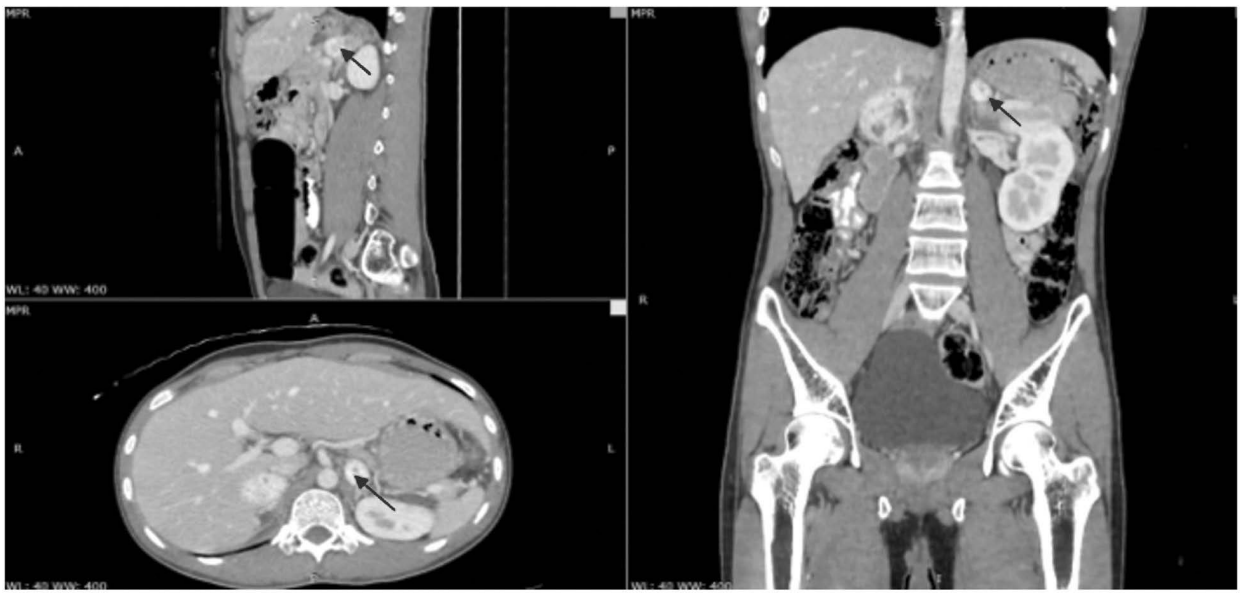

Figure 2. Left adrenal gland solitary tumor shown on different cuts from abdominal CT scan (arrows).

Treatment

After extensive discussions in multiple multidisciplinary team (MDT) meetings involving the Urology and Endocrine services, the patient underwent a total (open) adrenalectomy for the right adrenal gland, and an interval laparoscopic cortical sparing adrenalectomy for the left adrenal gland tumor after 4 weeks. A joint decision to perform an open adrenalectomy for the right side was established due to the size of the adrenal mass wherein the authors could not totally rule out a malignant process.

Laparoscopic cortical sparing adrenalectomy was performed transperitoneally and proceeded as standard practice similar to a laparoscopic total adrenalectomy as described by Stechman, 2022.[3] However, mobilization was minimized to just around the tumor, preservation of the adrenal vein was done, and only the tumor was excised, leaving normal adrenal tissue behind. The patient was placed in a left lateral decubitus position. Trocar placement was done as shown in Figure 3. The authors proceeded first with releasing the descending colon up to just distal to the left crus of the diaphragm. They ensured that the bowels and the tail of the pancreas would be deflected medially to avoid injury to surrounding structures. The Gerota's fascia was incised, and the main adrenal vein was identified. Dissection was continued superiorly to expose the superior aspect of the left adrenal along with the tumor (Figure 4). Once adequate exposure was achieved, the tumor was dissected off the normal adrenal gland with the use of ultrasonic shears, ensuring that a 5mm margin of normal tissue was included. Approximately 70% of normal adrenal tissue was preserved (Figure 5). After adequate hemostasis, the tumor was removed with the use of a sterile retrieval bag. Trocars were then removed and insertion sites were sutured. The patient had an unremarkable surgery. His vital signs were normal perioperatively, with no intraoperative adverse events encountered. In addition to this, no other pathologic lesions were seen in nearby organs and no blood transfusions were required. He was discharged 3 days after his surgery.

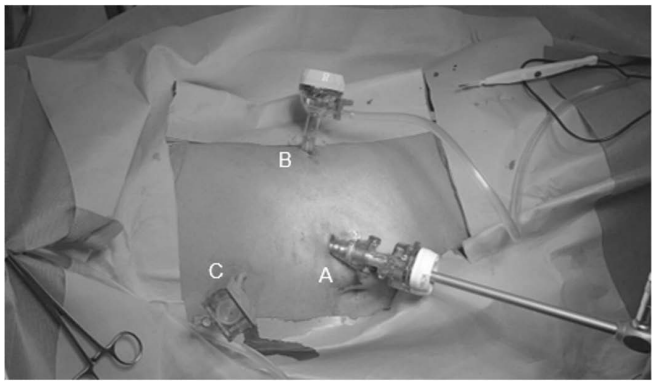

Figure 3. Patient positioning (left lateral decubitus) and trocar placement. (A) Visual port (12mm) was placed in the left periumbilical region, (B) 11mm trocar in the left subcostal region, and a (C) 5mm trocar in the subxiphoid region.

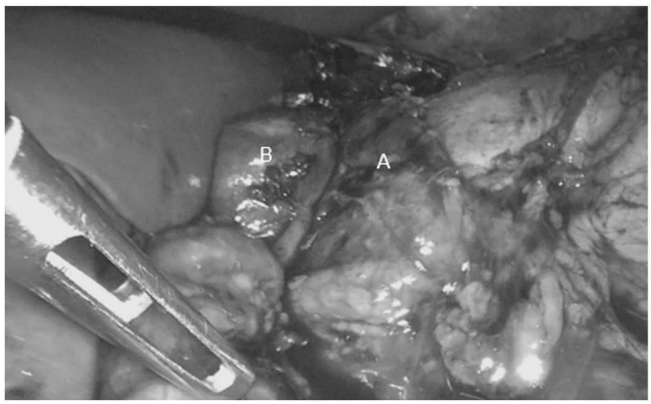

Figure 4. Exposure of the (A) left adrenal and the (B) tumor.

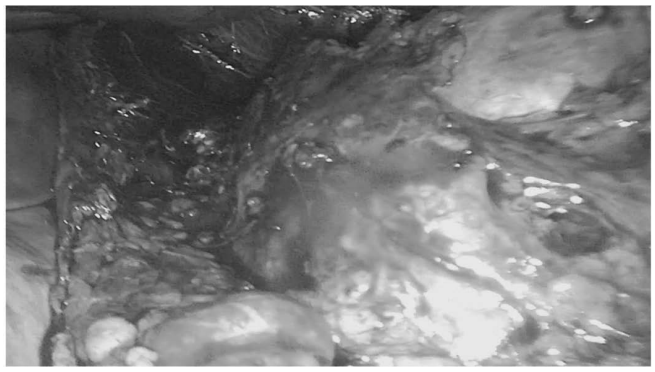

Figure 5. Left adrenal gland post-resection showing ~70% of residual normal adrenal tissue.

The pathology report for the right adrenal gland revealed pheochromocytoma, 5.0 centimeters in greatest dimension, with no lymphovascular invasion and negative tumor margins (Figure 6). The pathology report for the left adrenal gland revealed pheochromocytoma, 2.8 centimeters in greatest dimension, with no lympovascular invasion and negative tumor margins (Figure 7).

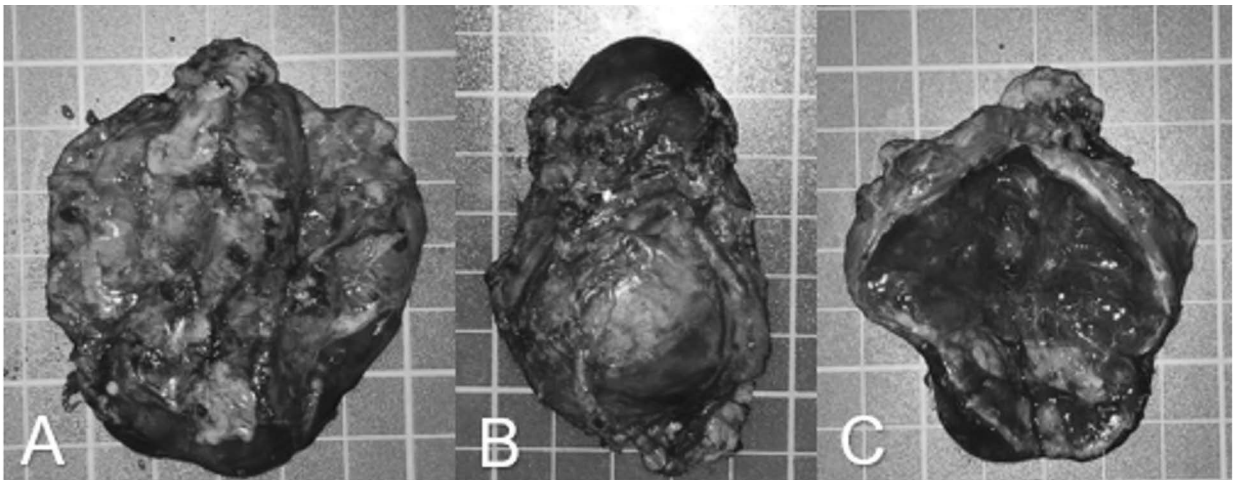

Figure 6. Right adrenal gland almost totally converted into a tumor (A) Anterior, (B) posterior, and (C) cut section views of the specimen.

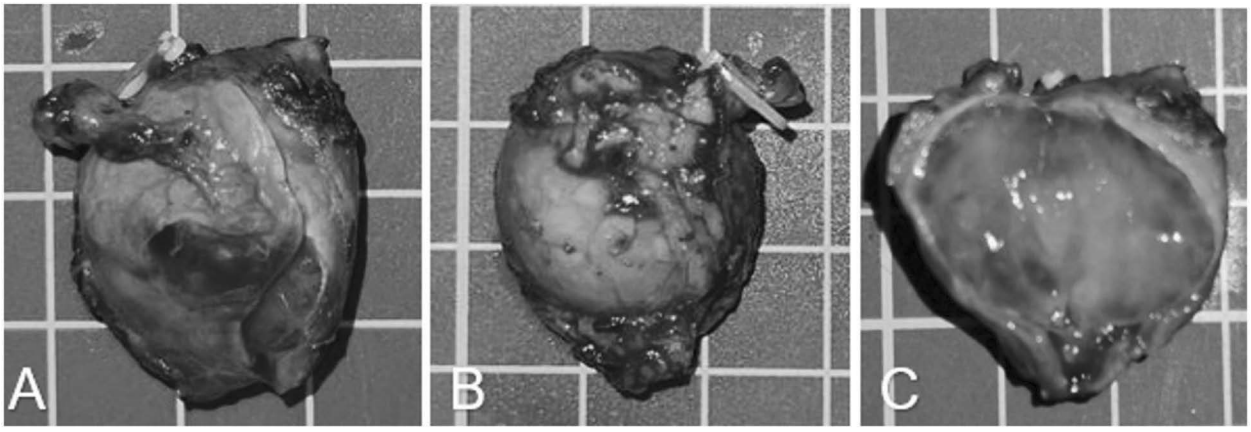

Figure 7. Left adrenal mass. (A) Anterior, (B) posterior, and (C) cut section views of the specimen.

He has been on constant follow-up with both the Urology and Endocrinology services and advised extensively regarding follow-up consults. He has had good blood pressure control since the surgery despite being off his anti-hypertensive medications and has had no symptoms of adrenal insufficiency despite not receiving any steroid replacement therapy.

Discussion

This report presents a rare case of vHL syndrome with bilateral adrenal tumors, managed with bilateral adrenalectomy in two separate surgeries wherein the laparoscopic cortical sparing adrenalectomy for the left adrenal tumor was the first ever performed at UP-PGH.

Cortical sparing adrenalectomy, which is also referred to as partial adrenalectomy or subtotal adrenalectomy, involves the resection of the pathologic lesion while leaving a significant amount of adrenal tissue. Two essential steps in performing a cortical sparing adrenalectomy are first, good exposure of as much of the adrenal gland without full mobilization, and second, preservation of its vascular structures.[4] A good margin of healthy adrenal tissue approximately 3 to 5 millimeters should be resected with the pathologic lesion. With regard to the amount of residual tissue that must be left behind after cortical sparing adrenalectomy, previous reports mentioned in a study by Perysinakis, et al. in 2020 recommend a volume of 15-30% in order to still be steroid-independent. Also mentioned was that if

a minimum of 15% residual adrenal tissue cannot be achieved, it is best to just perform an outright total adrenalectomy to decrease the chances of recurrence.[5] Cortical sparing adrenalectomy can be performed via open technique or via a minimally invasive approach. However, in recent times, the minimally invasive approach is preferred when feasible as it is proven to improve postoperative pain, shorten hospital admissions and hasten the recovery period.[4]

Cortical sparing adrenalectomy is recommended especially for patients with bilateral adrenal pathologies requiring surgical excision, to avoid the consequences of life-long steroid replacement which can lead to multiple complications if not monitored closely. During the early period of its conceptualization, surgeons initially would have to weigh the risks and benefits of performing a seemingly incomplete resection but with a patient safe from life-threatening Addisonian crisis. Some experts would also recommend cortical sparing adrenalectomy for unilateral tumors, attributing this to the fact that approximately 30% of patients are expected to develop contralateral adrenal disease over time hence the need to spare as much normal adrenal tissue as possible.[5] A review in 2010 by Kaye, et al. showed that cortical sparing adrenalectomy has a very low morbidity rate, very low recurrence rate (3%), and has approximately a 95% chance of granting patients freedom from steroid dependence.[6] Gumbs, et al. in 2006 specifically reviewed laparoscopic cortical sparing adrenalectomies mentioned in literature and found that mean operating times between total versus cortical sparing techniques did not differ significantly.[7]

In the setting of a lower middle-income country (LMIC) and that patient in the present study is from a rural area with fair access to healthcare, freedom from lifelong steroid replacement would greatly benefit him and his family as their funds can be reallocated to more basic needs. A retrospective study by Gunnarsson, et al. in 2017 explored the healthcare burden of patients with adrenal insufficiency from different etiologies. Aside from the fact that these patients must spend on the drugs, Gunnarsson, et al. found that patients suffering from adrenal insufficiency on steroid replacement had more frequent hospitalizations than patients in the control group. They concluded that irrespective of the cause of adrenal insufficiency, these patients had a significantly more substantial healthcare burden compared to their matched controls.[8]

This case of the first successfully performed minimally invasive cortical sparing adrenalectomy at UP-PGH, will pave the way for more cases to be performed using this technique. By doing so, surgeons can provide their patients the best quality of their care during their admission for their surgeries and at the same time, provide them with better quality of life, independent of exogenous steroids and the complications that come with it.

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CASE REPORT

Supine Endoscopically-Combined Intrarenal Surgery (ECIRS) for Encrusted Ureteral Stent with Staghorn Calculi, Ureterolithiasis and Cystolithiasis

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A forgotten and encrusted ureteral stent poses as a management dilemma especially when the encrustations are so severe that they involve the entire length of the ureteral stent. These can lead to staghorn formation, high volume ureterolithiasis and giant cystolithiasis which are all encasing the ureteral stent, This may lead to significant morbidity and mortality as a result of chronic urinary obstruction, recurrent urinary tract infection, and renal dysfunction and renal failure.

During the acute phase of the pandemic, a 31-year-old pregnant female, with 9 weeks age of gestation, underwent insertion of an indwelling ureteral stent for an obstructing renal pelvic calculus. She was lost to follow-up only to return two years later, with right flank and lower abdominal pains. Non-contrast CT showed encasement of the ureteral stent with a staghorn calculus on the proximal coil, extensive encrustations on the upper and middle segments, and a giant cystolithiasis at the distal coil of the ureteral stent. She underwent a supine endoscopically-combined intrarenal surgery (ECIRS), allowing retrograde retrieval of the ureteral stent after all the encrustations had been removed. There was minimal blood loss and no intraoperative and postoperative complications.

Encrusted ureteral stents with large stone burden may be treated effectively and safely with an endoscopically-combined intrarenal surgery. This combined antegrade and retrograde approaches to the urinary tract allows synchronous treatment of all calcifications around the forgotten ureteral stent, without resorting to open surgery.

Key words: encrusted ureteral stent, Endoscopically combined intrarenal surgery (ECIRS)

Introduction

An indwelling ureteral stent is usually inserted to drain an obstructed collecting system. It may be done acutely to recover renal function or as a means to empty infected urine, or as a terminal step at the consummation of an endourological procedure. It is not a permanent retention device and may not be retained for a protracted period. If forgotten and unremoved for a prolonged duration, its presence may lead to calcifications which may range from minor encrustations to large volume calculus formation which involve the proximal and distal coils or the entire length of the ureteral stent. This clinical condition is complicated by gross hematuria, obstruction, recurrent urinary tract infection, urosepsis, renal compromise and renal failure.[7]

Presented here is a case of a forgotten and encrusted ureteral stent which was managed successfully with a single-session supine endoscopically-combined intrarenal surgery

(ECIRS). The authors' experience in this case highlights that forgotten stents with high volume encrustations may be treated through a minimally invasive approach and should not necessarily be managed via open surgery. The authors describe their technique and the short-term outcome.

The Case

A 31-year-old female underwent emergent insertion of an indwelling ureteral stent for obstructive renal calculus during the acute phase of the COVID-19 pandemic. Two years later, she began to experience intermittent colicky right flank and hypogastric pains with a pain scale of 6-7/10, associated with urinary frequency, strangury and straining. There was no fever and gross hematuria. Unenhanced CT of the KUB revealed complete encasement of the proximal and distal ends of the indwelling ureteral stent by a 4cm x 3.5cm staghorn calculus (HU 1356) and a 5.2cm x 4.3cm cystolithiasis (HU 1356), respectively. There were also evident heavy calcifications in the upper and middle segments of the ureteral stent (Figure 1). After a thorough preoperative evaluation, she was given prophylactic antibiotic therapy with a third generation cephalosporin and was scheduled for supine ECIRS.

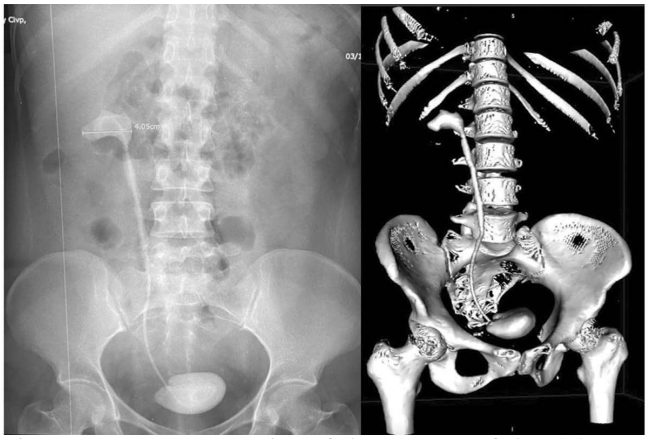

Figure 1. 3D reconstruction of the CT scan of the abdomen. Note the heavy calcifications surrounding both proximal and distal coils of the ureteral stent as well as the encasement of the ureteral segment of the stent.

Operative Technique

The patient was placed in a Galdakao-modified supine Valvidia position, with sandbags placed beneath the right scapular and pelvic areas respectively (Figure 2.) Fluoroscopic imaging confirmed the presence of radioopacities in the areas of the right kidney, the pelvic region, and along the course of the ureteral stent pertaining to the formation of staghorn, cystolithiasis and ureteral calculi, respectively.

Figure 2. Galdakao-modified supine Valvidia position. The patient was placed slightly laterally in Valvidia position, with the contralateral leg flexed.

The patient was prepared and draped in the usual sterile manner. A 36Fr nephroscope was then inserted transurethrally, visualizing a 60 cm ovoid cystolithiasis which formed around the distal end of the ureteral stent (Figure 3). This was fragmented with an ultrasonic lithotripter until it completely unraveled the distal loop of the ureteral stent. The bladder stones were evacuated completely using an Ellik bladder evacuator.

Upon visualizing the distal loop of the ureteral stent, the authors then proceeded to insert 0.889cm x 150cm Sensor® guidewire alongside it, until its tip was seen coiled within the renal collecting system. A 9.5Fr semi-rigid ureteroscope was advanced through the ureter, thus visualizing encrustations of the ureteral stent up to the renal pelvis. Ultrasonic intracorporeal lithotripsy was again utilized to clear the encrustations off the entire length of the ureteral stent. After completion, the guidewire was replaced with an open-ended ureteral catheter followed by retrograde pyelography, which revealed the staghorn calculus encasing the proximal coil (Figure 4).

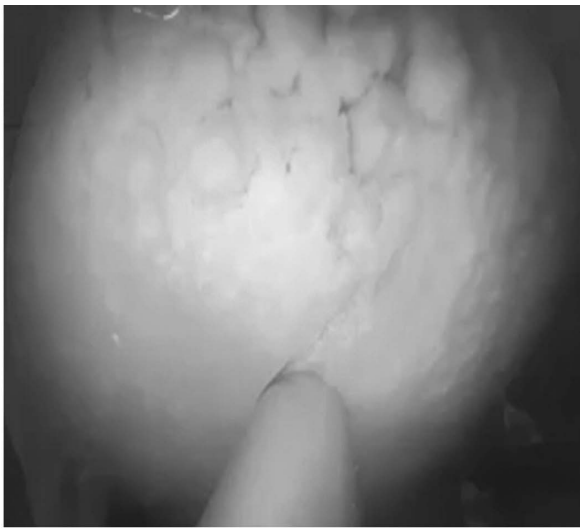

Figure 3. Cystolithiasis encasing the distal end of the ureteral stent pointed by the ultrasonic lithotripter.

Under ultrasound guidance (Figure 5), an 18G diamond-tip percutaneous renal access needle was introduced to enter the inferior calyx followed by antegrade placement of a Sensor® guidewire into the ureter and down the urinary bladder (Figure 6). This was then replaced with an Amplatz super stiff guidewire, followed by sequential renal tract dilation Amplatz fascial dilators up to 30Fr followed by insertion of the Amplatz sheath. A 26Fr nephroscope was introduced into the sheath to visualize the staghorn calculi which encased the proximal coil of the ureteral stent (Figure 7). Ultrasonic lithotripsy was again performed fragmenting the stones and detaching all of it from the ureteral stent (Figure 8). A tri-prong stone grasper was also used to evacuate all stone fragments. The ureteral stent was then extracted with ease from the urinary bladder using a 21Fr cystoscope. Two 16Fr Foley catheters were used as nephrostomy tube and bladder drainage.

Clinical Outcome

The total operative time was around 300 minutes and the estimated blood loss was minimal. The post operative course was unremarkable. Repeat KUB X-ray revealed no radio-opacity along the urinary tract (Figure 9.) The nephrostomy tube and the ureteral catheters were removed on post operative day. At one-month follow-up, repeat non-contrast CT KUB showed no residual stone fragments.

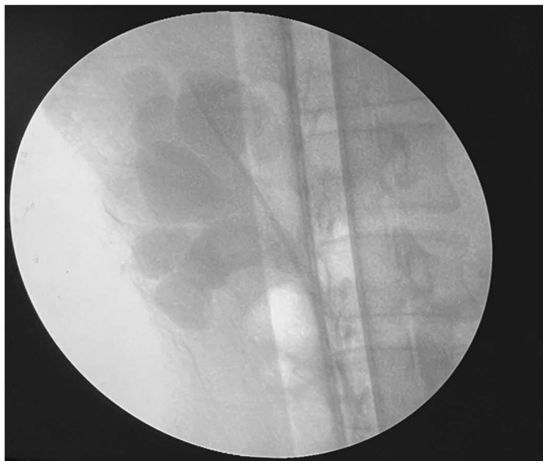

Figure 4. Open-ended ureteral stent was placed at the superior calyx followed by retrograde pyelography showing the staghorn calculus encrusting the proximal coil.

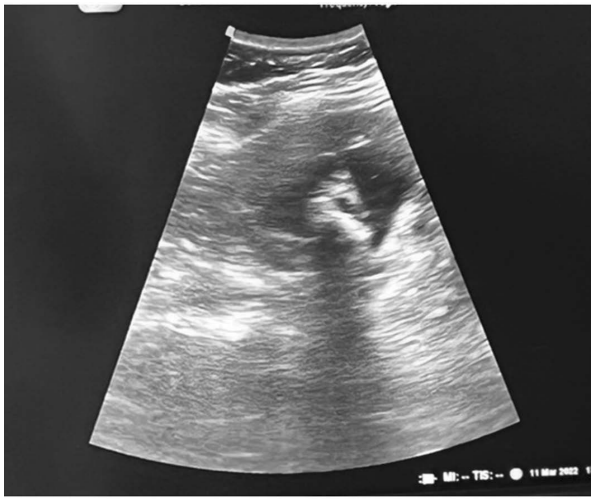

Figure 5. Kidney and Urinary Bladder Ultrasound showing a hyperechoic structure representing the staghorn calculi.

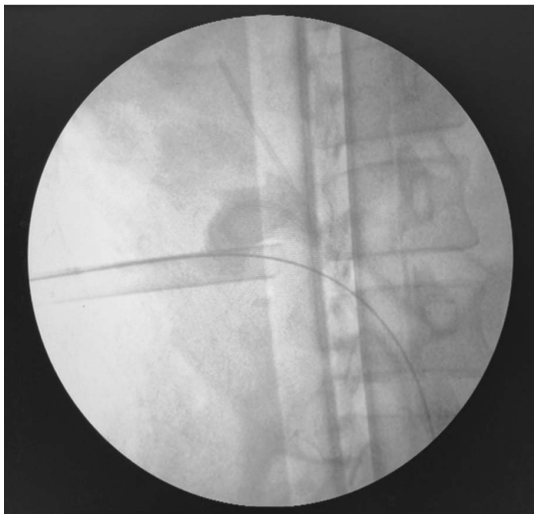

Figure 6. Antegrade placement of guidewire followed by renal tract dilatation and then insertion of Amplatz sheath.

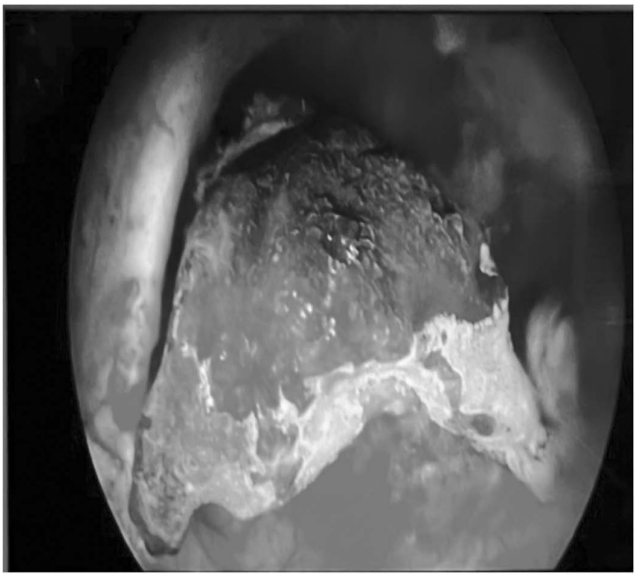

Figure 7. Staghorn calculi encasing the proximal end of the ureteral stent.

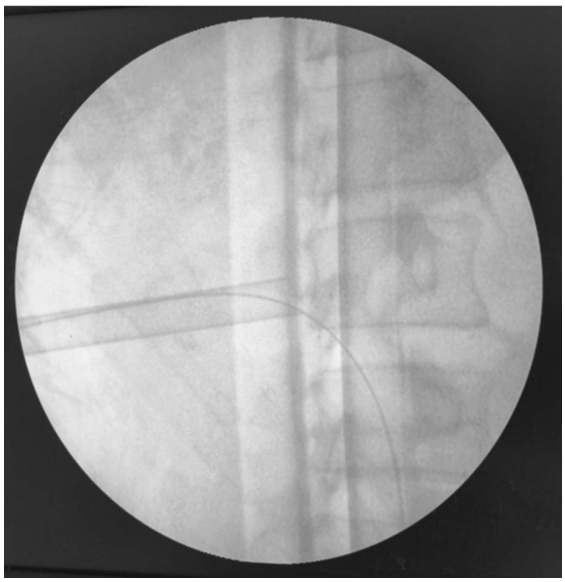

Figure 8. Repeat fluoroscopy after extraction of ureteral stent.

Figure 9. Repeat KUB X-ray showing absence of radio-opacity along the urinary tract, presence of Foley catheter balloon within the renal shadow and an open-ended ureteral stent.

Discussion

Forgotten and encrusted ureteral stents are serious health concerns. Considerable morbidity and mortality may occur as a result of gross hematuria, urinary tract obstruction, recurrent urinary tract infection, and renal compromise. The management options are variable and depend on the extent of calcifications. These may be monotherapy with, or a combination of, extracorporeal shock wave lithotripsy (ESWL), ureteroscopic intracorporeal lithotripsy (URS-ICL,) percutaneous nephrolithotripsy (PCNL) or open surgery. If a minimally invasive approach is favored, staged endourological procedures may be necessary to address the heavy stone burden that involves the bladder, ureter and the kidney. The choice of treatment is also influenced by the function of the affected kidney, the surgical expertise and the available technology.[9]

ESWL has played a role on encrusted stents depending on the severity and location of encrustation. Studies have shown successful treatment for proximal encrustations by ESWL monotherapy, while fully encrusted stent required multiple sessions of ESWL or open surgery. In this case, the authors utilized a single session of supine ECIRS. This allowed us to remove all the encrustations in a sequential manner using

an exclusively endoscopic approach, followed by ureteral stent removal, without the need for repositioning into the prone position. Ideally, different energy sources are required to achieve maximal stone clearance. The authors are limited only to the use of ultrasonic lithotripsy because of the unavailability of the laser which produces smaller fragments and less stone upward migration. Pneumatic lithotripsy is an option since it has shown to have shorter removal time, however, its stone fragments are too large to pass spontaneously and has a higher incidence of retropulsion to the kidney and retained stones. In another study, Recidoro et al described their experience with an encrusted stent which was managed with laser cystolithotripsy followed by a staged PCNL within the same hospital stay. The stent was removed intact from the percutaneous tract and the patient was rendered stone-free. The main reason for a staged approach was the prolonged OR time during the initial cystolithotrispy for a 7cm bladder stone. In contrast, in this current case report, the cystolithotrispy was facilitated with the use of an ultrasonic lithotripter which was introduced using a nephroscope. Fragmentation of stone using a semirigid ureteroscopy at the ureteropelvic junction was a challenge, considering the angulation from the inferior calyx. Preferably, a flexible ureteroscope would have easily bypass the angle in this area. The authors also did not see the need to delay the treatment of the staghorn component because the patient was stable. For this reason, there was no need to stage the PCNL on another occasion. The main advantage of these synchronous bladder, ureteral and renal endoscopic procedures is a single anesthesia experience and immediate recovery from the procedure.

While the authors were able to demonstrate the effectiveness and safety of supine ECIRS for the management of encrusted ureteral stents, they still believe that the best way to avoid devastating and catastrophic complications resulting from encrusted stents is really to avoid them from being forgotten. For this purpose, the patient should be well-informed of the potential dire consequence of a prolonged ureteral stent placement. On top of that, the health professionals should maintain a database for patients with indwelling stents so that they may be constantly reminded of their timely removal.

Conclusion

Supine ECIRS is a reasonable minimally invasive management of severely encrusted ureteral stents. The combination of both antegrade and retrograde approaches provide the opportunity to manage all the encrustations involving the entire length of the ureteral stent without the need for repositioning. The authors' experience show that it may be done effectively and safely to remove the stent while rendering the patient stone-free. Surgical expertise in endourology is required to undertake this challenging task.

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CASE REPORT

Vesico-utero-sigmoid Fistula Secondary to a Migrated Intra-uterine Contraceptive Device to the Urinary Bladder: A Rare Urogenital Complication

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Vesico-utero-sigmoid fistula secondary to an encrusted, transmigrated intrauterine contraceptive device (IUCD) to the urinary bladder is a rare urogenital occurrence. Reported here is a case of a 42-year-old female with 13 years of IUCD presenting with a two-year history of terminal dysuria, occasional hematuria and urinary dribbling. In the interim, she complained of persistent wet stools, pneumaturia, fecaluria and occasional urinary incontinence. Imaging revealed an encrusted IUCD with a concomitant vesico-utero-sigmoid fistula. Patient underwent a single setting colonoscopy, vagino-hysteroscopy, cystoscopy with cystostomy and extraction of encrusted foreign body (IUCD), excision and primary repair of vesico-utero-sigmoid fistula was done. The surgery proved successful, greatly improving the patient's quality of life. This is the first reported case of a vesico-utero-sigmoid fistula caused by a foreign body both in local and international literature.

Key words: Migrated intra-uterine contraceptive device (IUCD), vesico-uretero-sigmoid fistula, encrusted intrauterine device.

Introduction

Intrauterine contraceptive device (IUCD) is commonly used worldwide because of its reversible effects on female contraception.[1] Its safety, convenience, and low cost make it an appealing contraceptive option, especially in developing nations. Rare complications such as uterine embedment (IUCD located in the myometrium) and perforation (IUCD located beyond the uterine serosa) occur in approximately 1 in 1000 insertions.[2,3] Reported here is the first documented case of vesico-utero-sigmoid fistula secondary to an encrusted, migrated IUCD to the urinary bladder. This paper aimed to exhibit one of rarest complications of IUCD use, and the value of close follow-up after IUCD insertion. The authors would also like to emphasize the value of the different diagnostic modalities in the proper planning and management of vesico-utero-sigmoid fistulas.

The Case

The patient is a 42-year-old female, an overseas Filipino worker in Saudi Arabia, who came in the emergency room because of two-year history of terminal dysuria with occasional hematuria and urinary dribbling. Several consults were done in Saudi Arabia with a diagnosis of urinary tract infection. Kidney, ureter and urinary bladder (KUB) ultrasound revealed a migrated IUCD to the bladder. Patient was advised for surgical removal of the migrated IUCD, however, she opted to return to the Philippines for further intervention and was lost to follow up. One year prior to admission, persistence of symptoms, now with wet stools,

occasional pneumaturia, fecaluria and intermittent urinary incontinence prompted consult and referral to a urologist but with no compliance.

One month prior to admission, persistence of symptoms now with undocumented fever, dysuria and intermittency prompted urologic consult. KUB ultrasound and cystogram revealed an encrusted IUCD with leakage of contrast into the colon. The patient was advised admission but opted to transfer to this institution. Patient claimed that the IUCD had not been replaced or removed for 13 years.

On admission, genital, rectal and vaginal speculum exam were normal. Double-dye test was negative for fistulas. Urine GS/CS revealed growth of *E. coli* and was treated with Cefoxitin 1 g every 8 hours based on culture study. On pelvic X-ray, there was a T-shaped radio-opaque structure measuring approximately 3.7 cm in length with a round calcific density measuring 3.6 cm x 3.0 cm (Figure 1). The cystogram revealed adequate bladder distention and opacification of the urinary bladder lumen with extension into the colon (Figure 2). A contrast-enhanced abdominopelvic computed tomography (CT) scan confirmed a T-shaped radiopaque object with calcific densities (Hounsfield unit: 940-1050) measuring 3.1 cm x 2.6 cm inside the urinary bladder lumen. The delayed studies showed a fistulous tract in the postero-inferior aspect of the urinary bladder opacifying towards the sigmoid colon lumen (Figure 3). Given these following findings, a single setting colonoscopy, vagino-hysteroscopy, cystoscopy and open surgery was advised.

Colonoscopy was performed with urinary bladder irrigation of diluted methylene blue dye and fistulous tract was noted at the sigmoid colon, approximately 30 cm from the anal verge. An attempted cannulation of the fistulous tract was done; however, the tube can only be advanced approximately 0.5 cm (Figure 4). A vagino-hysteroscopy revealed an endometrial polyp at the posterior isthmus and a subsequent polypectomy was done. Multiple suspicious fistulous tracts at the anterior isthmus were noted, most probably from the previous area of IUCD migration. There was no egress of dye noted from the bladder (Figure 5). On cystoscopy, the urologists visualized an encrusted IUCD with approximately 1 cm solitary fistula located at the posterior wall of the urinary bladder (Figure 6).

Figure 1. Pelvic X-ray AP view: a T-shaped radio-opaque (arrow) structure with a round calcific density.

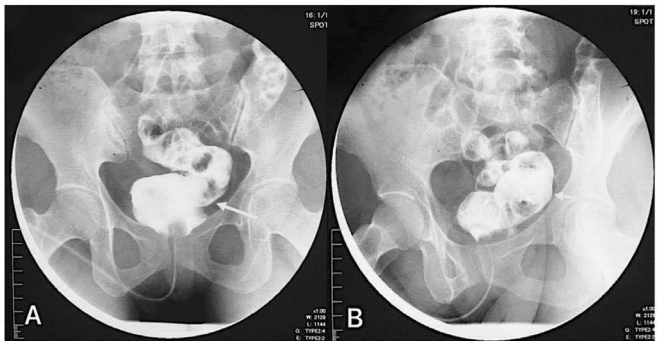

Figure 2. A – Cystogram AP view: opacification of the urinary bladder lumen with extension into the colon lumen (arrow); B – Cystogram Oblique view: opacification of the urinary bladder lumen with extension into the colon (arrow).

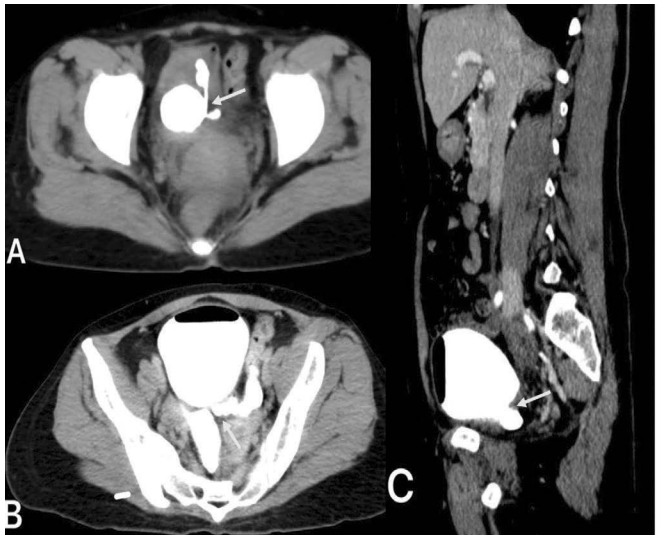

Figure 3. Abdominopelvic CT scan: A – Axial view: plain study showing a T-shaped radiopaque object with calcific densities (houndsfield unit: 940-1050) measuring 3.1 cm x 2.6 cm inside the urinary bladder lumen; B – Axial view; C – Sagittal view: both delayed studies showing a fistulous tract in the postero-inferior aspect of the bladder (arrow) opacifying to the sigmoid colon lumen.

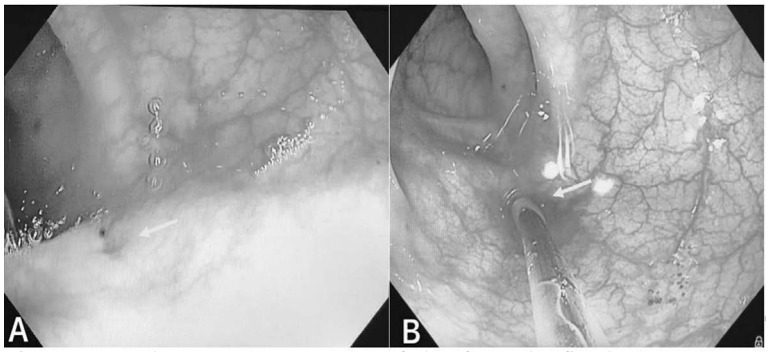

Figure 4. Colonoscopy: A –egress of dye from the fistula (arrow); B – attempted cannulation of the fistulous tract (arrow).

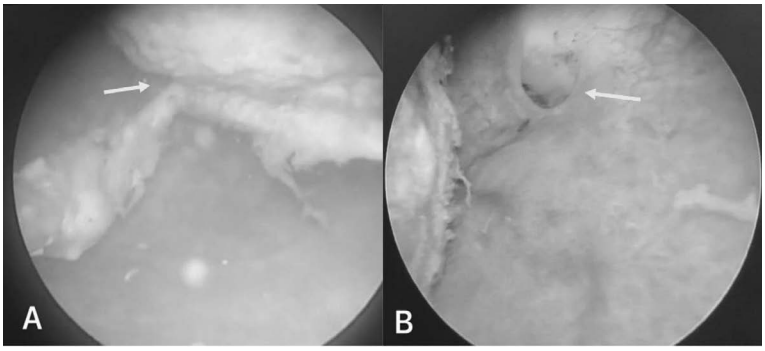

Figure 6. Cystoscopy: A – encrusted foreign body (arrow); B – fistula at the posterior bladder wall (arrow).

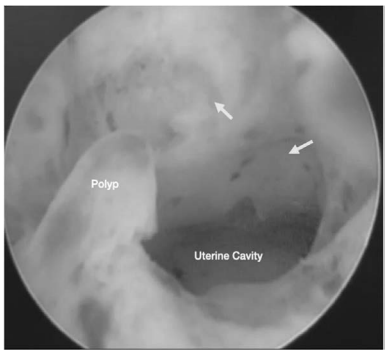

Figure 5. Vagino-hysteroscopy: endometrial polyp at the posterior isthmus with multiple suspicious fistulous tract at the anterior uterine wall (arrow).

Bladder exploration was performed through a vertical infra-umbilical approach showing the posterior area adherent to the sigmoid colon and the uterus where the fistula was located. A vertical incision was made approximately 1 cm below the fistula, anteriorly over the detrusor muscle up to the bladder mucosa exposing the encrusted, migrated IUCD (Figure 7). The migrated IUCD, containing approximately 4 cm oval-shaped encrustations, was extracted. A suspicious lesion was noted at the posterior bladder wall after the extraction of the encrusted IUCD (Figure 8) hence, sample tissues were taken for biopsy. The fistula was then cannulated with Fr 5 tube, noting a branched fistulous tract. One branch of the tract entered the sigmoid colon and the other branch entered the uterus. The fistula was then carefully excised separating the urinary bladder wall, sigmoid colon and anterior uterine wall (Figure 9). Debridement of the sigmoid colon and a double layer primary repair using continuous interlocking with Vicryl 3-0 suture for the first layer and interrupted with Silk 3-0 suture for the second layer was done. Debridement of the uterus and primary closure using continuous interlocking with Monocryl 0 suture was also performed. Finally, a Fr 18 foley catheter was inserted and the bladder defect was closed in a watertight, 2-layer repair using running Vicryl 3-0 suture for the mucosa and running Vicryl 2-0 suture for the muscularis. Bladder filling with 250 cc sterile water through the catheter was done with no note of leak.[6] An interpositional flap using the greater omentum was placed in between the posterior bladder and the uterus (Figure 10).

The post-operative course was uneventful and the patient was sent home on the fifth post-operative day with an indwelling foley catheter. Two weeks post-operatively, the repeat cystogram revealed an adequate bladder volume with no note of any leaks (Figure 11). The indwelling foley catheter was removed and the patient was able to void freely thereafter. After two months post-surgery, the patient was doing well, with no recurrence of symptoms. The histopathology report confirmed the diagnosis of a chronically inflamed fistula and the bladder likewise showed acute and chronic inflammation. The suspicious growth seen during hysteroscopy showed a benign endometrial

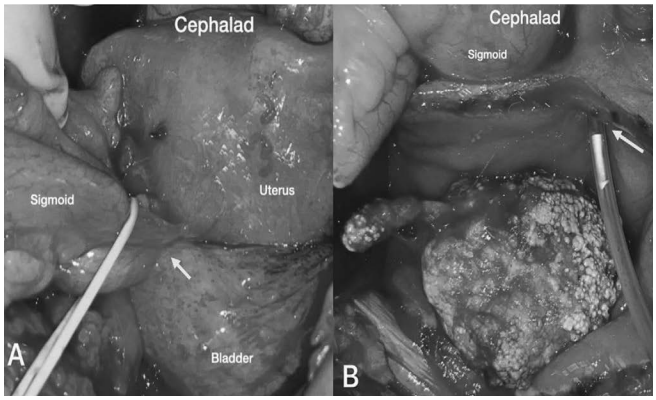

Figure 7. A – vesicosigmoid and vesicouterine fistula (arrow); B – bivalved urinary bladder with the encrusted foreign body (IUCD) inside; cannulated fistula at the posterior bladder wall (arrow).

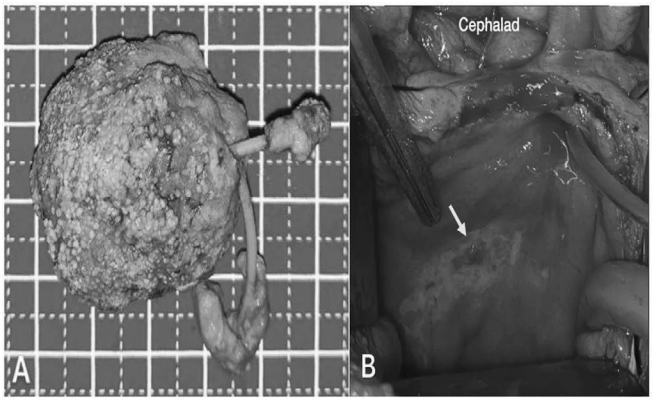

Figure 8. A – 4 x 3.5 cm encrusted foreign body (IUCD); B - bivalved bladder with suspicious lesion (arrow) noted at the posterior bladder wall after extraction of encrusted foreign body (IUCD).

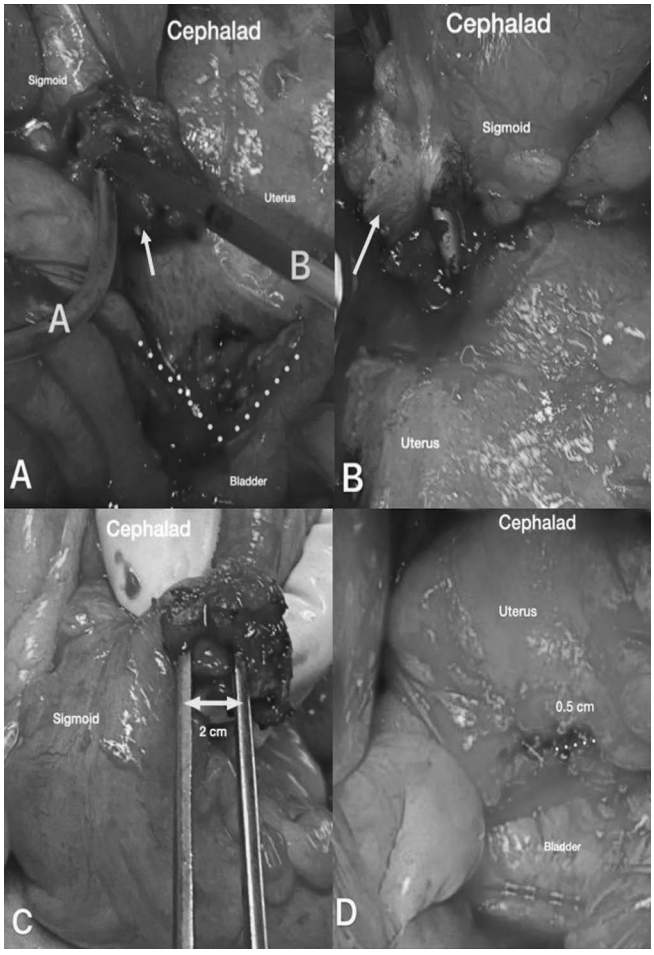

Figure 9. A – excised fistula (arrow) from the posterior bladder (dotted line) cannulated with 2 Fr5 tube showing the branched fistulous tract, one going to the sigmoid colon (A) and other going to the uterus (B); B – Vesicouterine fistula: excised fistula (arrow) from the bladder cannulated to the uterus; C – Vesicosigmoid Fistula: 2 cm in diameter (arrow), approximately 30 cm from the anal verge; D - Vesicouterine fistula: 0.5 cm in diameter (dotted line), anterior uterine wall.

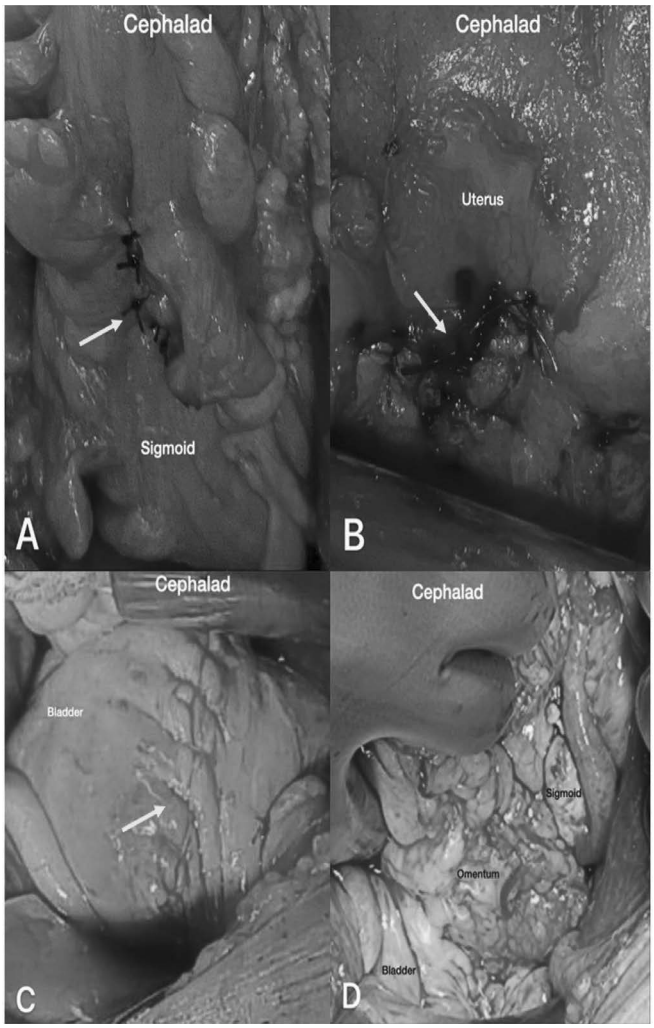

Figure 10. Debrided with primary repair vesicosigmoid fistula (arrow); B - Debrided with primary repair vesicouterine fistula (arrow); C – Cystorrhaphy (arrow); D – Interpositional flap (omentum) placed at the posterior bladder wall.

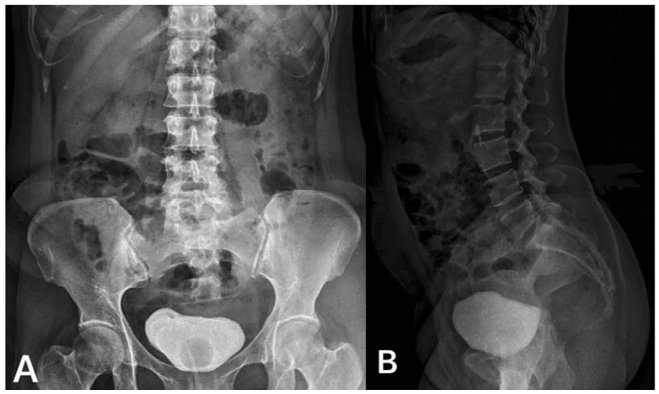

Figure 11. Cystogram: A – AP view: showing with adequate bladder volume without extravasation of contrast; B – Lateral view: no posterior extravasation of contrast.

polyp. The stone analysis of the encrustations revealed the composition of 58% carbonate apatite phosphate (dahllite), 30% calcium hydrogen phosphate dihydrate (brushite) and 12% magnesium ammonium phosphate (struvite).

Discussion

One of the rare complications of IUCD is migration. While cases are not large enough to warrant a statistical comparison, there is an increased risk if inserted immediately post-partum. Other risk factors for migration are use in nullipara, postabortion insertion, faulty technique of insertion, and irregular follow-up, as what happened in this case.[7,8] According to Joual et al, IUCD migration can be classified into incomplete or complete. Incomplete IUCD migration is seen when the device remains attached to the myometrium. Complete IUCD migration on the other hand is when the device drifts/travels to any site in the abdomen.[9] Incorrect direction to the uterine cavity, overestimation in the length of the uterine cavity, fragility of uterine wall due to recent birth, abortion, and pregnancy are contributory to the higher incidence of uterine perforation during IUCD insertion. After perforation of the uterine wall, IUCD can transmigrate to other adjacent organs such as the colon, wall of iliac vein, bladder, appendix, omentum, perirectal fat, retroperitoneal space, pouch of Douglas, ovaries, abdominal wall.[10,11] The authors deduce that iatrogeninc uterine perforation during IUCD insertion as the most plausible inciting factor in this case. Over the years, the intra-abdominal portion of the IUCD, particularly the arms, eroded and subsequently perforated the colon and the urinary bladder. With 13 years of neglect, proper placement of the IUCD was never assessed. Over time the intravesical portion of the IUCD, being exposed to urine, became a nidus for stone formation and growth. Slowly the encrustations grew leading to voiding symptoms. As time passed by the IUCD itself served as a plug hence patient remained asymptomatic. Bladder contractions must have slowly dislodged the IUCD towards the urinary bladder. Inflammation and fibrosis set in over the IUCD and sites of perforation, forming the fistulous tract. Once the IUCD has migrated, there has already a communication to the colon and uterus leading to the new onset of symptoms.

Acquired urinary tract fistulas are almost universally unexpected and may result in a great deal of inconvenience, discomfort, and physical disability to the affected individual. They are most often acquired because of a medical condition or surgical intervention for an unrelated problem. Vesicouterine fistula is a rare condition that only occurs in 1 to 4% of genitourinary fistulas. Gynecologic procedures such as low segment cesarean section are by far, the most common cause. It may or may not manifest with constant urinary incontinence because of the sphincter-like effect of the cervix.[12] Vesicouterine fistulas can be managed conservatively or through open surgery. Laparoscopic approach is feasible if done by an experienced surgeon.[13] Conservative management include prolonged indwelling bladder catheterization, fulguration of the fistula tract followed by hormonal induction of menopause have been used especially for small, immature fistulas. If these fail, the O'Conor transabdominal repair of vesicouterine fistula is the next option. The fistulous tract is excised from both structures, debridement of the uterus and bladder, and are closed individually with an interpositional flap, usually omentum, in between the two organs.[12] Vesicouterine fistulas that arise from a foreign body such as IUCD is also very rare. A case report by Szabó et al in 1992 described a 30-year-old female with urinary incontinence. The cystoscopic findings revealed an incompletely migrated, non-encrusted IUCD

perforated in the posterior bladder wall creating a vesicouterine fistula. The IUCD was removed cystoscopically through a grasper and initially managed conservatively with indwelling catheter for 6 weeks. Still with persistence of symptoms, they then did a transabdominal surgical approach. The surgery was successful, and the patient was discharged with improved symptoms.[14]

Uroenteric fistulas in general are most caused by diverticular disease (20%), Crohn disease (2-6%), and malignancy. Less common causes include radiation, infection, and trauma (external penetrating trauma, iatrogenic surgical trauma). Colovesical fistulas are commonly caused by diverticular disease in 75% of cases, with colon cancer, bladder cancer, radiotherapy, and Crohn's disease accounting for the remainder.[12] The standard approach for a colovesical fistula is an open surgery. Laparoscopic and robotic surgery is feasible and safe if done by an experienced surgeon. Non-surgical treatment is reserved to selected patients who are unfit for surgery. One-stage open surgical approach should be preferred, reserving the multi-stage procedure in patients with pelvic abscess, advanced malignancy or with previous radiation therapy.[15]

A combined colonic-gynecologic-urologic fistula is a much rarer condition. There is only one documented report presented as a case of a 74-year-old female with colo-vesico-vaginal fistula, however this was secondary to a sigmoid colon diverticulitis.[16] To date, there has been no reported case of a combined colonic-gynecologic-urologic fistulae that emerged from a foreign body specifically IUCD. The management of this case was based on the few case reports presented and following the basic principles of surgery for urinary tract fistulas.

In general, the principles of surgical management for urinary tract fistulas include: (a) adequate exposure of the fistulous tract with debridement of devitalized or necrotic tissue, (b) removal of involved foreign bodies or synthetic materials from region of fistula, (c) careful dissection and anatomic separation of the involved organ cavities, (d) watertight closure, (e) the use of well-vascularized and healthy tissue flaps for repair, (f) tension-free repair, (g) adequate urinary tract drainage after repair, and (h) prevention or treatment of infection.[12] Due to the complexity and rarity of what is causing the fistula, the authors decided that an open surgical procedure would be the best approach. In this approach, the urinary bladder, uterus, and sigmoid colon were clearly delineated. Through the cystostomy, the IUCD with a 4 cm encrustation was extracted. Meticulous inspection and debridement were done of the three involved organs and a watertight, tension free repair was ensured. Adequate urinary tract drainage after the repair was established through indwelling Foley catheter.

Conclusion

This case highlighted a very rare complication of a transmigrated IUCD. To the authors knowledge, this is the first documented case of vesico-utero-sigmoid fistula secondary to an encrusted, migrated IUCD to the urinary bladder. After IUCD insertion, regular follow up and examination are important to ensure its proper positioning and prevent IUCD transmigration to adjacent organs. Imaging modalities, combined with the help of video-assisted evaluations, and following the principles of surgical management of fistula would lead to a successful long-term outcome. Lastly, this case foregrounded that even in the advent of endo-laparoscopic and robotic urologic surgery, open surgery still proves to be an integral part of the urologist's armamentarium.

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