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**BCG therapy for high-risk NMIBC:  
A national referral hospital experience**

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**Laparoscopic Pyelolithotomy -  
another tool for large renal stones**

# Kenya Journal of Urology

The Kenya Journal of Urology (KJU) will be published quarterly and the next issue is expected to be published in September 2022. The Editor-in-Chief is calling for manuscripts for consideration. All manuscripts submitted will undergo peer review so it's advisable to submit them in good time.

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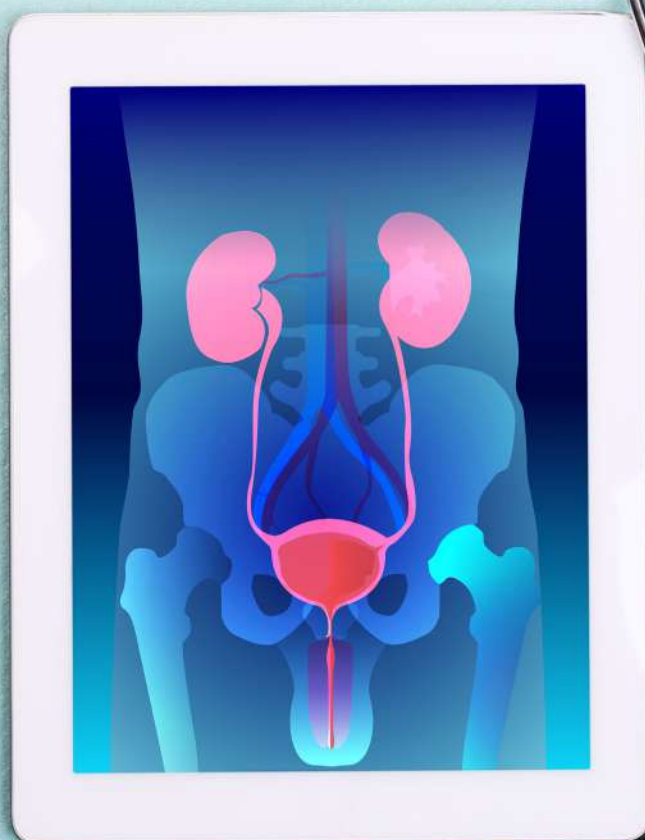
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## Editor's note

This is the second issue of the Kenya Journal of Urology. The nine peer-reviewed articles in this issue are based on research in urology done mostly in Kenya both adult and paediatric populations.

The journal accepts articles from researchers in the field of urology. This publication shows how urology has now established itself as a speciality in the East African region.

A lot of the research is clinical work in the region that may provide information on decision making in patients who need urology care ranging from the undescended testis through testicular torsion to bladder cancer, urological laparoscopy. Case reports on surgical approaches to a number of pathologies provide insights to newer approaches in urology.

A third issue of the journal will provide continued change in the art and science of urology with a regional emphasis.

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Prof. Peter Mungai Ngugi  
**Editor-in-Chief**

# The landscape of urological research in Kenya

## Osawa F

MBChB, MMed, FCS, Consultant Paediatric Surgeon and urologist, Lecturer department of Surgery, University Of Nairobi, P.O. Box 19676 Nairobi, Email:osawaf@uonbi.ac.ke

Research is crucial in health care. Translation of in-clinic or in-lab discoveries into practical interventions that reduce illness burden and enhance the quality of life for target populations is a key objective of biomedical research.

These objectives are frequently accomplished through the widespread distribution of knowledge derived from high-quality research published in peer-reviewed publications, which has an impact on clinical practice. The Kenya Journal of Urology (KJU) acts as a conduit for the dissemination of peer-reviewed research results among Kenyan, regional, and even international health professionals.

On the African continent, the visibility of urological sciences is low. Similar to other countries, Kenya has a low level of urology research production, as seen by the publications in both national and international urology journals. Changing this narrative and offering a venue for the publication of local and regional research work are among the objectives of KJU. There will be sufficient data for study in all areas of paediatric and adult urology with the commissioning of the East Africa Kidney Institute in Nairobi.

Paediatric urological conditions with adequate data available for research locally include congenital and acquired genital abnormalities, urinary tract disorders, urooncology, and renal disorders among others. Research in aspects of sexuality and fertility for instance in relation to congenital and acquired genital abnormalities is covered in this issue. The article by Said et al highlights a crucial component of assessing testicular development and function by assessing testicular volume in children with undescended testis. The importance of salvaging a testis that has undergone testicular torsion to preserve fertility by the use of a reliable scoring system that is time-saving has been emphasized by Otieno et al in this publication.

This issue has also highlighted some aspects in the spectrum of pediatric sexual medicine in conjunction with commonplace pediatric urological conditions. The article by Mustafa et al addresses trends and outcomes of reconstructive surgical procedures in the paediatric population previously managed for hypospadias which certainly has a bearing on some aspects of sexuality and fertility.

In rare cases of penile trauma, penile reconstruction to an extent of allowing for adequate sexual function is necessary. Phalloplasties are the last resort for these boys. A case report by Mustafa et al in this issue has brought to light one of the rare causes of penile trauma by a constrictive object and the potential danger that the penis is exposed to during the removal of such objects.

Knowledge gained from research, increasing experience, new technological advances, and non-invasive diagnostic screening modalities have had a profound influence on treatment modalities in adult urology in Kenya, a trend that is likely to continue in the years to come. A practical and preliminary approach to selected adult urological conditions is outlined in this issue of the KJU. The article by Aideed et al on laparoscopic pyelolithotomy highlights this progress in the practice of adult urology in Kenya. The work by Kimani and Kortimai et al on bladder cancer is certainly a great move towards the addition to the body of knowledge in urooncology in Kenya. However, like in paediatric urology, there is very little if any research work in sexual and fertility medicine despite the abundance of data.

Finally, I want to express my gratitude to the editorial board, the authors, and the publishers for their work on this issue of KJU. These articles should inspire and encourage readers to conduct urology-related research and publish their findings in upcoming issues of this journal.

# Assesment of testicular volume at age of presentation in patients with unilateral undescended testes as seen at Kenyatta National Hospital

Said M<sup>1</sup>, Osawa F<sup>1</sup>

<sup>1</sup>University Of Nairobi

**Corresponding Author:** Dr F O Osawa. Email: osawaf@uonbi.ac.ke

## ABSTRACT

**Background:** Undescended testes (UDT) is known to affect testicular function, as seen by the quality of spermatogenesis, testosterone levels, and semen profile. Testicular size serves as a good indicator of testicular function. A crucial component of assessing testicular development and function is testicular volume assessment in undescended testis.

**Objective:** The aim of this study was to evaluate testicular volume upon presentation in paediatric patients with unilateral undescended testes seen at Kenyatta National Hospital.

**Method:** This was a prospective cross sectional study conducted between November 2021 and May 2022 of paediatric patients with unilateral undescended testis seen from the Pediatric Surgical Outpatient Clinic of Kenyatta National Hospital whose demographic data were collected and testicular volume determined radiologically.

**Results:** The mean age was 5.02 years. The majority (85.7%) of the participants were born at term and the mean birth weight was 2.9 kgs. The volume of the undescended testis was smaller than the descended side and this was statistically significance above the age of 2 years ( $p < 0.001$ ).

**Conclusion:** The significant difference between the volume of undescended and descended testis in this study may be an indication that the testicular function of an undescended testis after the age of 2 years could be affected.



## INTRODUCTION

Undescended Testes (UDT) is one of the most frequent anomaly of male genitalia with incidence of 2 to 5% of term male neonate (1,2). The incidence of unilateral undescended testes is about 90% and bilateral is about 10%(1,6). There is risk of testicular malignancy about 3-8 folds in the retained testicles(1,2).

Testicular function is affected by UDT. Studies have shown that spermatogenesis is disrupted in the gonads with abdominal or inguinal locations (5,10). The quality of spermatogenesis, testosterone levels and semen profile are reflected by the testicular size (9,12). The testicular volume assessment in undescended testis is a key aspect to evaluate the testicular development and function(6).

Late presentation and higher locations are associated with significant decrease in testicular volume (6,10). The size of the UDT is smaller markedly compared to the descended one preoperatively (9,11,12). Also no significant differences have been found in testicular volume between cryptochid and healthy children below the age of 6 months (12,13,15). This study was seeking to assess testicular volume at age of presentation in patients with unilateral undescended testis and hence help in the determination of the optimal timing for surgery.

### Materials and methods:

This was a prospective cross-sectional study carried out at KNH Pediatric surgical outpatient clinic and radiology department between November 2021 and May 2022. The study population were male infants and children with unilateral UDT at the age of 6 months and above who were recruited consecutively. Infants and children with other genitourinary anomalies like hypospadias or those who had received hormonal therapy were excluded.

Sample size estimation of 78 participants was calculated using statistical Fishers formula with a Standard deviation of the 95th percentile (1.96) and a confidence interval of 95%. The expected proportion of patients with undescended testes was calculated out of the number of total operations per year done at the Paediatric Surgical Unit at Kenyatta National Hospital and which was 5.4%.

Informed consent and Biodata was obtained from the parent or guardian of the participants. The Biodata included age of child in months or years ,age at first presentation ,Weight in Kilograms Height in centimeters and birth history. A physical examination was then conducted to confirm the diagnosis of undescended testes and determine the laterality. Testicular dimensions were measured at the radiology department using PHILIPS ultrasound probe C8-5 Microconvex and UPP-110 Sony printer from which the volume was calculated. All the information obtained was entered into a data collection tool.

Data were entered into a Statistical Product and Service Solutions (SPSS) version 26 programme for analysis. Descriptive statistics such as means, medians and mode were used to describe characteristics of the study participants. Results were presented in written reports, frequency tables, bar and pie charts and line graphs. Continuous variables were compared using Mann-Whitney U test. Correlations were tested using Spearman rank test. A p value of less than 0.05 was considered statistically significant

## RESULTS

### Age:

Mean age of the 70 participants whose age was recorded was 5.0 years, SD 3.39, Median 4, Range 0.5 years – 13 years. Only 9 participants (13.0%) presented below the age of 2 years.

70% of children who presented with left UDT were 4 years old and below, while 47.5% of children who presented with right UDT were 4 years and below.

Figure 1: Histogram of Age distribution in patients presenting with cryptorchidism

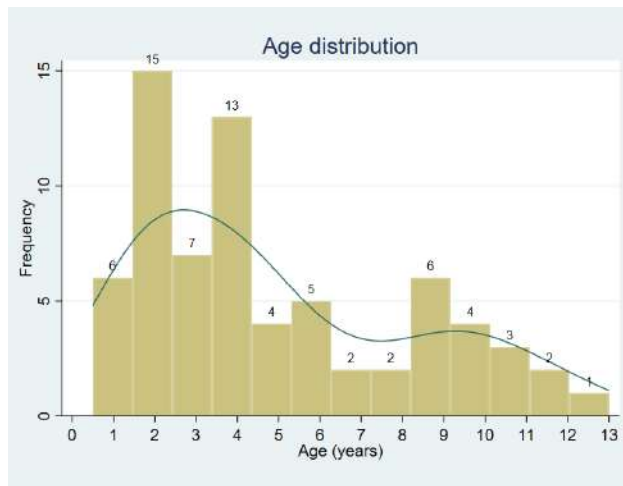


Table 1: Comparison of age of presentation and side of cryptorchidism

	Right UDT	Left UDT
<= 4 years	19	21
>4 years	21	9

**Gestation at birth**

Majority of the participants were term by gestation (85.7%, n=60). The distribution by gestation of birth is summarized in figure 3 below

**Birth weight**

The mean birthweight 2.9 kgs, SD =0.66; Median 3 kgs, Range 0.99 – 4 kg

**Laterality of undescended testes**

Right UDT (n=40) occurred more frequently than the left (n=30)

**Testicular volume**

Table 2: Testicular volume in left undescended testes

Testicular volume	N	Mean	SD	Median	Range	P value
Left UDT	30	0.625	0.226	0.685	0.18 – 1.04	0.103
Right descended testes	30	0.69	0.252	0.71	0.25 – 1.31	

P value derived from Mann- whitney u test

In left UDT, the mean testicular volume at presentation in the left and right testes were 0.625 and 0.69 respectively. This association was not statistically significant with a P value of 0.103.

Table 2 shows the testicular volume variable in left UDT

Table 3: Testicular volume in right undescended testes

Testicular volume	N	Mean	SD	Median	Range	P value
Right UDT	40	0.575	0.017	0.615	0.38 – 0.79	0.001
Left descended testes	40	0.709	0.199	0.705	0.5 – 1.27	

P value derived from Mann- whitney u test

In right UDT, the mean testicular volume at presentation in right and left testes were 0.575 and 0.709 respectively. This association is statistically significant with a P value of 0.01.

Figure 2: Scatterplot of testicular volume vs age in left cryptorchidism

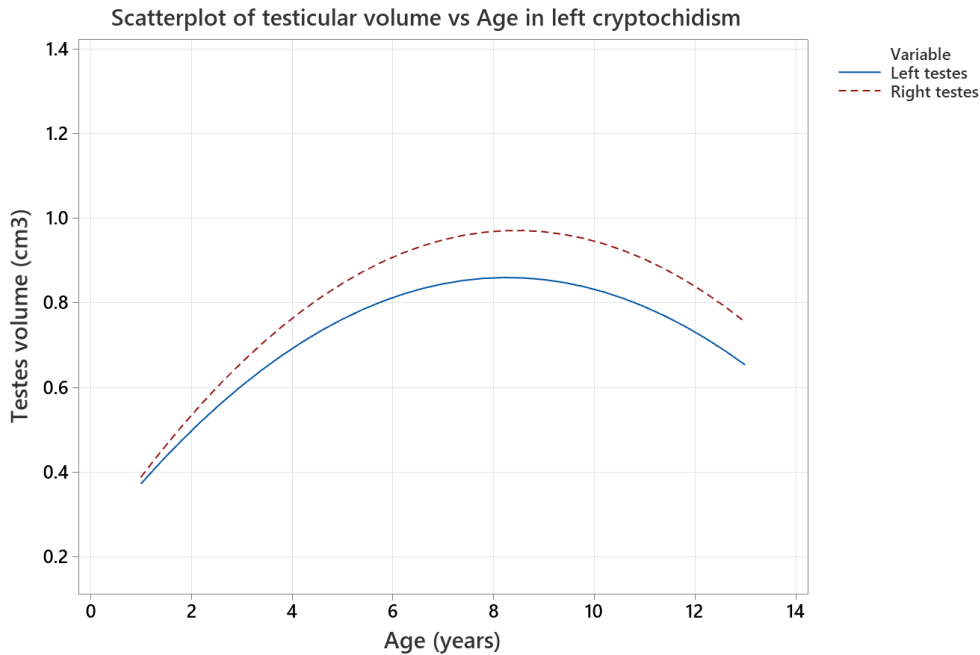


Figure 3: Scatterplot of testicular volume vs age in right cryptorchidism



There was a significant correlation between increase in age and testicular volume in both left and right unilateral UDT. This correlation is noted to be more significant in the right UDT, where the volume steadily lags behind the descended counterpart with increasing age.

Notably the changes in testicular volume is seen to occur with significant variability at 2 years of age in both left UDT (P value 0.048) and right UDT (P value 0.02).

## DISCUSSION

The quality of spermatogenesis and testosterone levels are affected by testicular volume. Children with UDT had been noted to have smaller testicular size and less sperm concentration. In the literature, late presentation is associated with decrease in testicular volume. However, significant difference in testicular volume was not reported between cryptorchid and healthy boys younger than 6 months of age. This study sought to assess testicular volume at age of presentation in patients with unilateral undescended testis and hence the optimal timing for surgery.

### Age presentation

In this study, the mean age of the 70 participants whose age was recorded was 5.0 years, SD 3.39, Median 4, Range 0.5 years – 13 years. Only 9 participants (13.0%) presented below the age of 2 years. Thus, delayed presentation is a common phenomenon in this population.

A study by Ekwunife et al (2018) in Nigeria noted that only 9 (23.1%) of patients presenting before 2 years. Numerous other studies have shown a late presentation as a common feature in the African setting (39). The study by Ekwunife et al identified the risk factors for late presentation as ignorance by parents on the abnormal position of the testes, wrong advice by clinicians during first visit, and misguided belief that testes will descend (39). This study did not investigate the factors associated with late presentation in our set up.

### Gestational age and birth weight

Cryptorchidism is generally increased in preterm compared to term neonates. The prevalence at full term is 2 – 5% which further decreases to 1 – 2% at three months. Amongst the preterm the incidence increases to up to 45% (22). In our study, Majority of the participants were term by gestation (85.7%, n=60). The mean birthweight 2.9 kgs, SD =0.66; Median 3 kgs, Range 0.99 – 4kg. Low birth weight (<2.5kgs) has been considered as a risk factor for cryptorchidism (41). Other notable risk factors include maternal smoking and diabetes (5).

### Laterality of UDT

Right UDT (57.1%, n=40) occurred more frequently than the left (42.9%, n=30). This data is consistent with reports in the literature which shows that 60% of cryptorchidism occurs on the right side compared to 20% in left side and bilaterality in 10%. A study by Ekwunife et al (2018) in Nigeria noted similar results of laterality 41% (16 patients out of 39 cases) (39). While 54.8% patients had right sided UDT in study by Steven Kadilo et al in Tanzania (1)

### Testicular volume

Participants with left UDT, mean testicular volume at presentation in left and right testes were 0.625 and 0.69 respectively. This association was not statistically significant with a P value of 0.103. Unlike the participants with right UDT where the mean testicular volume at presentation in right and left testes were 0.575 and 0.709 respectively. This association is statistically significant with a P value of 0.01. This is explained by the fact that in this study only 30% of children who presented with left UDT were older than 4 years of age, while 52.5% of children who presented with right UDT were older than 4 years of age. These findings are consistent with studies done by Hutson et al and Mathers et al who reported that late presentation are associated with significant decrease in testicular volume (6,10).

Also Our study showed that there was a significant correlation between increase in age and testicular volume in both left and right unilateral UDT. This correlation is noted to be more significant in right UDT where the volume steadily lags behind the descended counterpart with increasing age. Notably, the changes in testicular volume is seen to occur with significant variability at 2 years of age in both left UDT (P value 0.048) and right UDT (P value 0.02).

## CONCLUSION

This study showed a considerable difference between the volume of undescended testes and descended testis, with the difference becoming more significant after 2 years of age. This could be a sign that testicular function after the age of two years may be compromised. Therefore, it is best to have surgery before the age of two. According to the study's findings, cryptorchidism frequently presents late in Kenyan society. Therefore, public health initiatives should aim at raising knowledge of parental and clinical testicular examination in newborns and infants to facilitate early management among children with cryptorchidism.

## Authors contribution

Both the authors (Dr Said M, Dr F Osawa) participated in the design of the study and the interpretation of data. The manuscript was drafted by Dr F Osawa. Both the authors critically revised the manuscript and have approved the final version for publication.

## Conflict of interest

The authors declare no conflict of interest

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# Trends in urethrocutaneous fistula management post hypospadias surgery at Kenyatta National Hospital

**Mustafa Omar<sup>1</sup>, Francis Osawa<sup>1</sup>**

<sup>1</sup>Department of Surgery, University Of Nairobi

**Corresponding Author:** Dr F O Osawa. Email:osawaf@uonbi.ac.ke

## ABSTRACT

**Background:** Urethrocutaneous fistulas are common complications that occur after hypospadias surgery. The development of such fistulas is dependent on the severity of hypospadias, surgical technique, and experience of the operating surgeon. Many methods have been developed for UCF correction, and the best technique for fistula repair is determined based on the size, location, and number of fistulas, as well as the status of the surrounding skin.

**Objective:** The aim of this study was to establish the trends in urethrocutaneous fistula surgery at the Kenyatta National Hospital (KNH)

**Method:** Data from records of patients who had undergone urethrocutaneous fistula surgery between January 2017 to 2021 December were collected and analyzed retrospectively. The data included details regarding the type of hypospadias repair done, the timing of fistula development, duration before the first attempt at repair, intraoperative details, postoperative management, and outcome of the repair.

**Result:** Proximal hypospadias had the highest fistula rate of 58% and developed very early after urethroplasty. Most repairs (62%) were performed after a 6 months period from the time of fistula development. The preferred suture material was PDS (69%) and a majority of stents or catheters (67%) were left in place for 7-14 days postoperatively. The overall success rate was 87% after the first attempt at fistula repair.

**Conclusion:** Repair of UCF that occurs after hypospadias surgery at KNH is mostly done at least 6 months after its appearance. Polydiaxone (PDS) is the suture material of choice for repair. The favored practice is to leave a catheter or stent after the repair for 7-14 days. These protocols appear to have good outcomes with a success rate of 87%.

**Keywords:** Urethrocutaneous fistula, Hypospadias

## INTRODUCTION

Urethrocutaneous fistulas that occur after hypospadias repair are common complications not only locally but also globally. The majority of fistulae occur in the immediate postoperative period, and this has been supported by a study using porcine models (1) Based on the severity of hypospadias, surgical technique, and experience of the operating surgeon, the incidence of UCF ranges from 5% to 10% (2).

Jumbi et al in a study done at KNH found a high incidence of 47% for urethrocutaneous fistulas post hypospadias repair, which obviates the need for further insight into this complication of hypospadias surgery (3). This study also established that wound infection and meatal stenosis were the most significant factors associated with the development of urethrocutaneous fistula post hypospadias repair.

Despite the existence of certain basic principles for the management of urethrocutaneous fistulas, Paediatric urologists have a few differing preferences for managing different types of fistulas. The aim of this study was to evaluate the trends in the management of urethrocutaneous fistulas that developed following hypospadias surgery at Kenyatta National Hospital and to establish how these practices contribute to the overall outcome.

## METHODS

This study was a retrospective review of all patients who underwent hypospadias surgery repair at KNH, a tertiary teaching hospital in Nairobi, Kenya, from January 2017 to December 2021. All patients who developed urethrocutaneous fistulas as a complication were included in the study. Records that were incomplete were excluded from the study. Perioperative details that were extracted from the records included the type of hypospadias that necessitated the repair, time interval for the appearance of the urethrocutaneous fistula after the hypospadias repair, duration between the appearance of UCF and first attempt at repair, type of repair done, type of suture used, whether a catheter or stent was left in situ after the repair and if so for how long and finally the outcome of the repair.

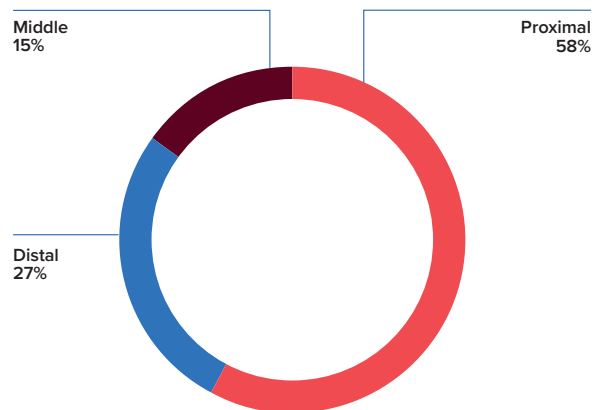
Descriptive data analysis was done and the results were presented in percentages, pie charts, and bar graphs.

## RESULTS

During the study period, 210 hypospadias surgeries were performed at KNH but only 150 records were retrieved and from which 26 were of patients who developed urethrocutaneous fistulas as a complication. The perioperative management of these patients was analyzed in detail.

Fifty-eight percent (58%) of urethrocutaneous fistulas occurred following repair for proximal hypospadias, followed by distal hypospadias at 27% and midshaft hypospadias at 15%.

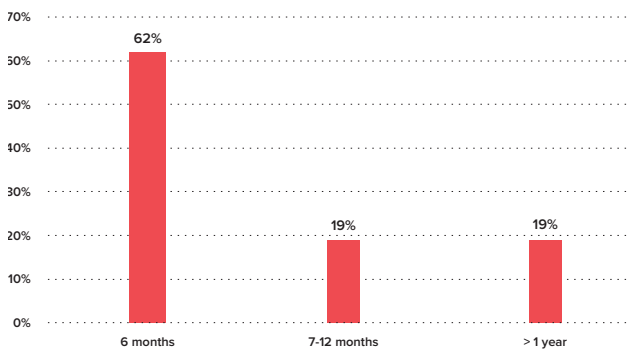
Figure 1: **Distribution of hypospadias by type at the time of initial urethroplasty.**



Sixty-eight percent (68%) of urethrocutaneous fistulas developed immediately after removal of the catheter post-operatively while 14% were noticed 2-4 weeks following surgery.

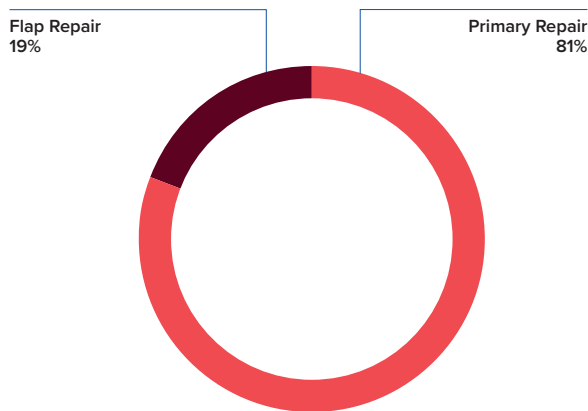
The majority of the repairs (62%) were done 6 months from the time of development of the UCF. A sizable number of the repairs (19%) were done between 7 months to one year. Those who had repairs done beyond one year were initially lost to follow One patient presented with UCF five years after urethroplasty for hypospadias.

Figure 2: Time interval between UCF development and repair



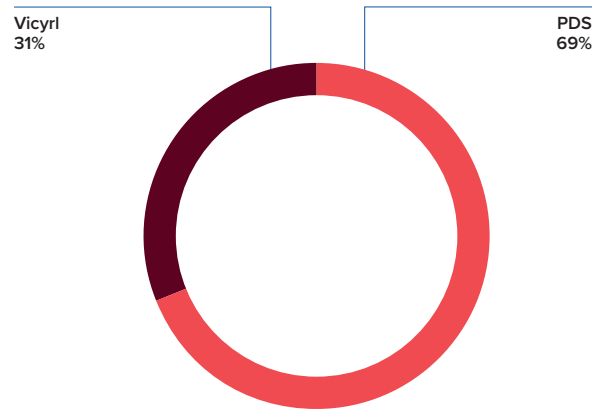
Simple closure which entails multilayered repair with a waterproofing layer was done in 81 percent of the patients at the time of primary repair of the UCF. The use of local skin flap surrounded was the procedure of choice in 19% of the patients. There was no records on the indication for the choice of procedure done.

Figure 3: Type of repair



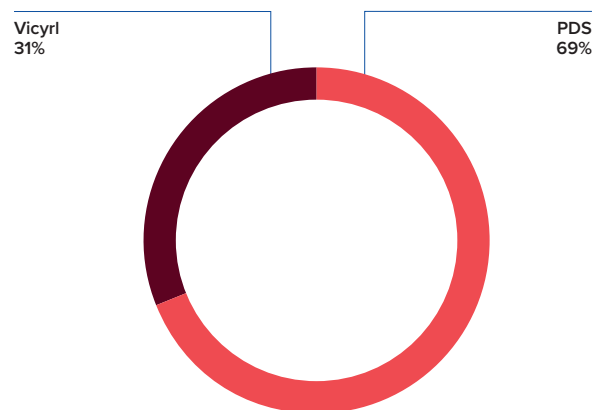
As for the type of suture material, the synthetic absorbable monofilament suture polydioxane (PDS) was used in 69% of cases and synthetic braided suture Vicryl was used in 31% of the cases. There was no statistically significant difference in the outcome based on the type of suture used.

Figure 4: Type of suture



Following UCF surgery in this study, a catheter or stent was left in place for each patient. The catheter or stent was left in place for 7 to 14 days in 67% of patients, 7 days or less in 25%, and more than 14 days in 8% of instances. The longest time frame was 21 days.

Figure 5: Duration of catheterization or stenting after UCF repair



Regardless of the type of procedure done, the suture material used or the duration of stenting or catheterization the overall success rate in this study was 87%. This is in keeping with global success rates which range from 80-91% after the first attempt at repair of UCF post urethroplasty for hypospadias.



## DISCUSSION

The goal of repairing hypospadias is correct chordee and locate the urethral opening at the tip of the glans to achieve not only an aesthetically pleasing organ but also optimize its sexual and urologic function. (3) The mechanism of UCF development lies in the incorporation of urethral mucosa or neo-urethra in ventral repair with rapid migration of urethral mucosa and skin epithelium into suture tracts, usually due to infection, ischemia, or both (4) (5). Urethrocutaneous fistulas are a common complication of this repair and hamper the initial goal of the surgery. It is therefore important to identify this complication and deal with it and get back on track in achieving the original goal.

Most urethrocutaneous fistulas occurred after the repair of proximal hypospadias. This is in keeping with global data and could be attributed to the complexity of repair of these types of anomalies with expected complications such as infection and meatal stenosis that usually leads to the development of these fistulas. (7)

At KNH, most of the UCF were first noticed at the time of catheter removal after the initial hypospadias repair. Most reported fistulas occurred within the first month postoperatively and from the study this was the same finding since 82% of the UCF appeared within the first month after repair.

After identifying the presence of a UCF, the patients are followed up in the outpatient clinic, and repair is attempted after a minimum of 6 months. This is a common practice globally since this period allows tissues to settle, inflammation to subside and the tissues to heal completely before attempting a repair of the fistula. At KNH, most surgeons (62%) were found to prefer to wait for six months before attempting a repair. Some favored up to one year waiting period.

The most effective choice for fistula repair depends on the size, location, and number of the fistulas as well as the condition of the skin around them. Several techniques have been devised for UCF correction. Simple closures can easily treat smaller fistulas, but if the fistula is bigger and surrounded by vascularized skin, a local skin flap is required (7). In this study 81% of cases had a simple repair of UCF was the preferred procedure. This entails locating the fistula, removing the fistulous tract, initially closing the fistula, and then covering with a layer of skin and dartos. This produce less complications and recurrences. Although the multilayer closure procedure has a low recurrence rate, it takes a long

time to learn, is challenging, and there is a chance that the UCF will enlarge during the tract excision. Flap repair was done in 19% of the patients. In this study, there were no records to suggest the reason for the choice of flap repair procedure however, in literature, large fistulae >2mm and multiple fistulae could be an indication for choosing this type of repair (9).

Gentle tissue handling, urethral mucosa inversion following excision of the epithelialized fistula tract, multilayer repair with a waterproofing layer, non-overlapping sutures, the use of absorbable and thin suture materials, a tension-free closure, the use of optical magnification, and needle-point cautery for coagulation were some of the principles for the ideal repair technique described in a previous studies (10). Information regarding some of these principles was not available in the records used as the source of data in this study

Polydioxane (PDS), a synthetic, absorbable monofilament suture, was the most frequently utilized suture for repair in this study, being used in 69% of the cases. However, vicryl, a synthetic, absorbable braided suture was employed in 31% of the cases. There was no clear explanation in the records for why these sutures were selected in these circumstances. Regardless of the type of suture utilized, there was likewise no statistically significant difference in the results. A catheter or stent was left in place after surgery for every patient at KNH. The duration, though, was different. While 25% only kept the catheter in for less than a week, the majority of surgeons (67%) preferred to leave the catheter or stent in for 7 to 14 days. Just 8% of surgeons preferred to leave the catheters in for more than two weeks.

All of these procedures resulted in an overall success rate of 87% for UCF repair on the first attempt at KNH following hypospadias surgery. This is comparable to worldwide data, which show an 85–96% success rate. (9)

## CONCLUSION

At Kenyatta National Hospital, UCF that develops following hypospadias surgery is often repaired at least six months after it manifests. Primary repair is preferred, and PDS is the preferred suture material. The recommended procedure is to keep a catheter or stent in place for 7–14 days following the repair. The 87% success rate of these techniques suggests that they have positive results.

### Authors contribution

The study's design and data analysis were done with input from both authors (Drs. O Mustafa and F Osawa). Dr. F. Osawa was the author of the manuscript. The final version of the text has been authorized for publication after being critically revised by both authors.

### Conflict of interest

The authors declare no conflict of interest.

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# Evaluation and validation of BAL testicular torsion scores in a cohort of children with acute scrotum at Kenyatta National Hospital

Otieno C<sup>1</sup> Osawa F<sup>1</sup>

<sup>1</sup>University of Nairobi

**Corresponding Author:** Dr F O Osawa. Email: osawaf@uonbi.ac.ke

## ABSTRACT

**Background:** Testicular torsion is one of the common causes of acute scrotum and it requires immediate intervention to prevent testicular loss and infertility. There is significant clinical overlap among the various causes of acute scrotum and this has led to the development of different scoring systems like the BAL score among others to aid in the early diagnosis of testicular torsion.

**Objective:** The aim of this study is to evaluate and validate the BAL testicular torsion score in a cohort of children with acute scrotum at the Kenyatta National Hospital in Nairobi, Kenya

**Method:** Data was collected retrospectively for all male children under the age of 15 years who had been diagnosed with acute scrotum in the period between 2013 and 2021. The BAL score was then calculated and the presence or absence of other patient characteristics were also recorded.

**Results:** A total of 34 patients were included out of which 26 patients had testicular torsion. Sixteen patients had complete records and were included in the final analysis. The sensitivity of the BAL score was 64%, specificity was 100%, Positive predictive value was 100% and the Negative predictive value was 55%.

**Conclusion:** The BAL score is a reliable predictor of testicular torsion, and it should only be used in conjunction with ultrasonographic analysis to increase diagnostic precision without delaying the therapy of patients that may otherwise be saved by the score alone.

## INTRODUCTION

Acute scrotum is defined as “the constellation of new-onset pain, swelling, and/or tenderness of the intrascrotal contents. “Some of the causes of acute scrotum include epididymo-orchitis, testicular appendage torsion, testicular torsion, (90%); idiopathic scrotal edema, varicocele, tumor, strangulated inguinoscrotal hernia and trauma (1)

Testicular torsion is defined as a rotation of the longitudinal axis of the spermatic cord resulting in strangulation of testicular blood flow. It is a surgical emergency and prompt intervention is warranted to prevent testicular loss and impaired fertility. One in every 4000 males under the age of 25 years develops testicular torsion annually (2). The peak incidences of testicular torsion have been reported to be between 12 and 14 years (more in <18 yrs) by some studies with a smaller peak in the neonatal age group. It accounts for only a small fraction of acute scrotum cases in children (10–15%) and exploration in all children would result in unnecessary surgery in most cases (3). Therefore, a fast and definite diagnosis is essential (4)

Diagnosis may be challenging as most reasons for acute scrotum show a high clinical overlap and imaging may not be readily available (5, 6)

### Testicular salvage rates

The time to presentation remains the most important factor in predicting outcomes in children with acute testicular torsion. Early intervention (within 6 hours) results in a testicular salvage rate of 90% - 100%, and this rate decreases after 6 - 12 hours to 20%-50%, and after 12 - 24 hours to 0 -10% (7). Locally the salvage rate has been reported to be low at 21% with an orchidectomy rate of 79% (8). Teenage testicular torsion occurs intra vaginally and is associated with a significantly higher salvage rate as opposed to Neonatal torsion that is usually extra vaginal and has a poor salvage rate of around 9%.(9,10)

### Testicular torsion scores

Traditionally the diagnosis of testicular torsion relied heavily on clinical presentation but there exists a considerable clinical overlap among the various causes of acute scrotum. For this reason, various scores have been designed to improve diagnostic accuracy and especially when these scores are combined with imaging findings (11)

Some of the scores include Testicular Work up for Ischemia and Suspected Torsion (TWIST) score , Boettcher Alert (BAL) score, Artificial Intelligence (AI) score

Doppler ultrasonography combined with clinical presentation provides an excellent sensitivity of 85% - 100% and specificity of 75% -100 %. (1 2)

The BAL-Score in particular has enhanced specificity in comparison to the other scores. This score is very easy and quick to access. It includes only four features, which are direct to access via medical history and physical examination, so no time delay is expected. The components of the score include a duration of pain < 24 h, a high riding testicle, nausea and/or vomiting, and absent cremasteric reflex. Each component is given a score of one (13,14)

## STUDY OBJECTIVE

To evaluate and validate the BAL testicular torsion score in diagnosing testicular torsion in a cohort of children with acute scrotum at Kenyatta National Hospital

## MATERIALS AND METHODS

This was a retrospective study and involved a review of patients' records in the period between 2013 and 2021. All male paediatric patients under the age of 15 years who had a diagnosis of acute scrotum at Kenyatta National Hospital and met the inclusion criteria were included in the study. Records of patients who could not express their symptoms (neonates), patients who had reduced perception of symptoms for testicular torsion (spinal cord injury below T10), patients who had sustained testicular trauma, and patients with incomplete records ( $\geq 50\%$  of components of BAL Score) were excluded from the study. Consecutive nonrandom sampling was used to enroll patients who met inclusion criteria

Data on patient characteristics were abstracted from the records and recorded in a predetermined data collection tool. The Patient characteristics included; Duration of pain, Nausea/Vomiting, Position of the testicle, Presence of cremasteric reflex, Presence of scrotal erythema, Fever, Dysuria, Tenderness of testicle/epididymis, Blue dot sign, Serum markers of infection (Full Haemogram, C-Reactive Protein), Urine analysis, Microbiological findings (microscopy/culture/sensitive ty), Findings on ultrasound, Intra-operative findings.

Each of the 13 patient characteristics (predictor variables) was evaluated in all the children with acute scrotum and their presence or absence was noted and utilized to calculate the BAL score.

The main outcome variable was the presence or absence of testicular torsion. The definitive diagnosis of absence or presence of torsion was confirmed from the records of surgical exploration by examination of the testes.

The data was analyzed using Statistical software for social sciences(SPSS). Chi-square analysis was then conducted for each of the 13 predictor variables to determine their association with testicular torsion. Sensitivity, specificity, Positive predictive value, and negative predictive value of BAL score for prediction of testicular torsion were determined.

## RESULTS

### Testicular torsion rate

A total of 34 patient records were examined in the period between 2013 and 2021.

Out of 34 patients' records, 26 had testicular torsion translating to a testicular torsion rate of approximately 76% in patients with acute scrotum.

Over 10 years old made up the majority (52.9%) of patients with acute scrotum. Similarly, 85.3% of patients were seen at the hospital 24 hours after the onset of symptoms.

Figure 1: **Age distribution (acute scrotum)**

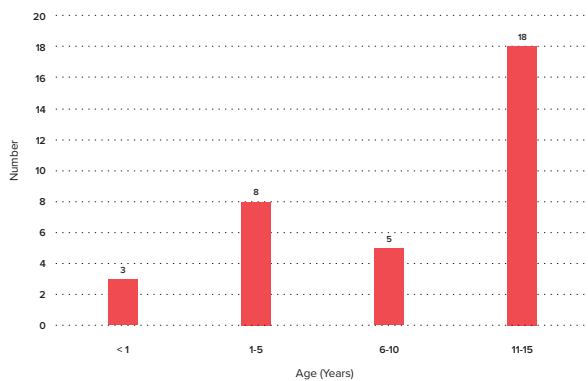
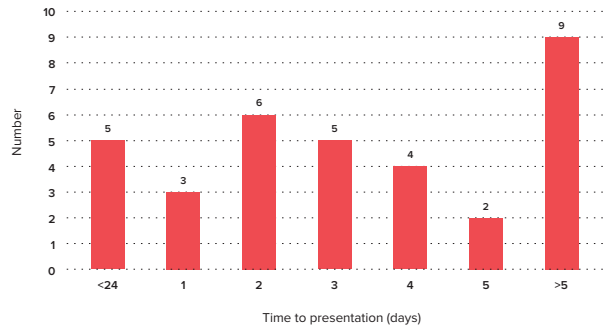


Figure 2: **Time to presentation (acute scrotum)**



### BAL score analysis

A total of 18 patients out of the 34 patients whose records were examined were excluded in the final analysis of the BAL score due to incomplete records.

Patients with records missing 2 or more components of the BAL score were excluded from the final analysis. Fifteen out of the 18 patients who were excluded had testicular torsion and 3 did not have testicular torsion.

Seven patients had complete records of all the components contained in the BAL score. A total of 16 patients had 3 or 4(all) components in the BAL score and were included in the final analysis.

Among the components of the BAL score, cremasteric reflex had the highest number of missing records(24/34).

Table 1: **BAL score of the study participants**

NUMBER OF PATIENTS	YES			NO			N/A	P-VALUE
	Testicular Torsion +	Testicular Torsion -	Total	Testicular Torsion +	Testicular Torsion -	Total		
Duration of pain < 24 hrs	5	0	5	20	8	28	1	
Nausea/Vomiting	11	1	12	6	7	13	9	
High riding testicle	11	0	11	1	5	6	17	
Abnormal cremasteric reflex	5	0	5	2	3	5	24	
Fever (>37.5)	0	0	0	21	6	27	7	
Dysuria	1	0	1	5	5	10	23	
Presence of scrotal erythema	3	1	4	4	4	8	22	
Tenderness of testicle/ Epididymis	22	5	27	1	2	3	4	
Blue dot sign							34	
Serum markers of infection (WBC>11,CRP>4)	2	2	4	22	6	28	2	
Urine analysis (L+++ , N+)	0	2	2	3	2	5	27	
Urine m/c/s (m+/c+)							34	
Ultrasound findings (↓ / - blood flow on doppler)	16	1	17	5	6	11	6	

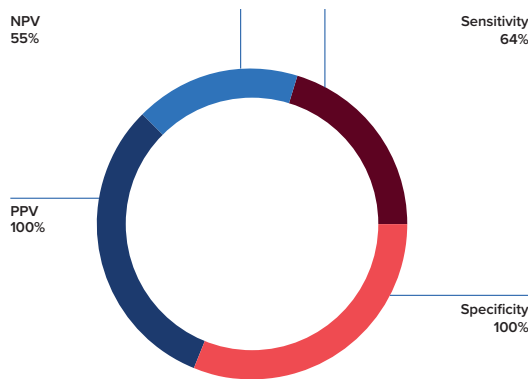
Table 2: **BAL score values**

BAL SCORE	Testicular Torsion +	Testicular Torsion -	TOTAL
0	2	5	7
1	2	0	2
2	3	0	3
3	4	0	4
4	0	0	0
<b>TOTAL</b>	<b>11</b>	<b>5</b>	<b>16</b>

Table 3: **BAL score evaluation**

SENSITIVITY/SPECIFICITY/NPV			
BAL SCORE	TESTICULAR TORSION		TOTAL
	PRESENT	ABSENT	
	7	0	7
	4	5	9
<b>TOTAL</b>	<b>11</b>	<b>5</b>	<b>16</b>

Figure 3: **BAL score sensitivity, specificity and predictive values**



## DISCUSSION

Acute scrotal conditions are common in children and present with scrotal pain, swelling, and redness in the affected hemiscrotum. The true cause is difficult to determine. There are myriad etiologies for this syndrome, including torsion of the testis (TT), torsion of the testicular appendix (TTA), epididymo-orchitis (EO), and strangulated inguinoscrotal hernia (SIH)(15,16,). Keiichiro et al found that acute scrotum in 23% of their patients was due to testicular torsion (17). In this study, however, testicular torsion was found to be the cause of acute scrotum in 76% of all the cases, a rate that is much higher than that quoted in many studies at less than 40%. The explanation for this is beyond the scope of this study but could be due to the under-diagnosis of the other aetiologies.

The peak incidence of testicular torsion in most literature is in the pubertal age group (12–17 years) (18,19) which is consistent with the findings of this study. The number of patients between the ages of 11 and 15 formed the majority (52.9%) of the cases.

Testicular torsion being a surgical emergency warrants immediate intervention to prevent testicular loss and subsequent infertility. There exists a significant clinical overlap among the various causes of acute scrotum and this can significantly delay the diagnosis of testicular torsion. In addition to its duration, the degree of rotation has been implicated in the clinical outcome.(20,21) Ischemia can occur as soon as 4 hours after torsion and is almost certain after 24 hours. (22). In this study 85.3% presented to the hospital more than 24 hours after the onset of symptoms with ischemic testes and had to undergo orchidectomy. The high disease burden of testicular torsion as the major cause of

acute scrotum and the significant delays in presentation with subsequent loss of testes calls for the adoption of a quick and accurate diagnostic tool to improve the outcomes.

The BAL score is an excellent predictive tool for testicular torsion in children with acute scrotum and can enable early diagnosis. In the current study, the BAL score was noted to have a sensitivity of 64% and a specificity of 100%. This study also established that the BAL score has a negative predictive value of 100% and a positive predictive value of 55%, a confirmation of its reliability as a predictive tool for testicular torsion. Out of 11 patients, 7 would have been accurately identified by the BAL score as having testicular torsion and 4 would have been missed.

## Study limitations

This being a retrospective study missing data limited the analysis of the BAL SCORE and other predictor variables in finding their association with testicular torsion.

The records of 17 patients were incomplete and were not analyzed.

Inguinoscrotal hernia as a cause of acute scrotum but was included as a variable in the current study due to limitations of the study design as it was not possible to differentiate purely inguinal hernias from inguinoscrotal hernias from the available records. Given that Inguinoscrotal hernias constitute approximately 1.7% of cases of acute scrotum locally (and less than 10% in literature) its inclusion may not have significantly altered the final results.

## CONCLUSION

Testicular torsion as a surgical emergency that requires immediate intervention necessitates the need for a quick diagnosis. The BAL score is a reliable predictive tool for testicular torsion in children with acute scrotum. Combining the BAL score with ultrasonography does improve the accuracy of diagnosis of testicular torsion however, sonographic evaluation should not delay surgical exploration in cases that can potentially be salvaged.

## Authors contribution

Drs. C. Otieno and F. Osawa, who are also authors, took part in the study's design and data interpretation. Dr. F. Osawa was the author of the manuscript. The final version of the text has been authorized for publication after being critically revised by both authors.

## Conflict of interest

The authors declare no conflict of interest

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# Factors affecting the low uptake of radical cystectomy in Muscle Invasive Bladder Cancer (T2-T4a) disease at a Kenya National Referral Hospital

L.B. Kortimai<sup>1</sup>, F. A. Owillah<sup>1</sup>, J. Ikol<sup>2</sup>, P. Mburugu<sup>2</sup>, D. Kimani<sup>2</sup>

<sup>1</sup>Department of Surgery, University of Nairobi

<sup>2</sup>Department of surgery, Kenyatta National Hospital

**Corresponding Author:** Dr L.B. Kortimai. Email: kortimairejoice@gmail.com

## ABSTRACT

**Background:** Radical Cystectomy and Urinary diversion are the standard surgical treatment for muscle-invasive bladder cancer (MIBC), and there is a Low reported uptake of treatment for Muscle Invasive Bladder Cancer. Although most guidelines propose radical Cystectomy as the mainstay of treatment for MIBC, there is minimal uptake of radical Cystectomy and urinary diversion in Kenya.

**Objective:** The aim of this study was to determine the factors that affect the uptake of radical Cystectomy at the Kenyatta National Referral Hospital in Nairobi Kenya.

**Method:** This was a 5-Year retrospective census study from 2017-2021 in which data was collected from records of patients using a structured questionnaires and online questionnaires from service providers (Urologists). The data collected which included: Patients related data, Hospital data on surgeon volume and surgeon perception on radical Cystectomy were analyzed for factors that affect the uptake of radical Cystectomy.

**Result:** Out of 56 patients who had Muscle Invasive Bladder Cancer, only 27 (48%) had complete data that were analyzed. Majority of the patients (59.3%) who had radical cystectomy were below 66 years old compared to 7.4% of the patients with advanced age (80years and above). Registration with insurance (NHIF) was a significant favorable factor for radical Cystectomy at 55%. In 55.6% who had no reported comorbidity only 37% had radical Cystectomy. A significant proportion of patients (55.6%) opted for trimodal therapy. Those who declined intervention or lost to follow-up were 7.4%. All the urologist involved in this study had done less than 2-5 cases of radical cystectomy per year (low volume).

**Conclusion:** Despite a relatively younger population with fewer comorbidities which favors the criteria for radical Cystectomy, patient's choice for Trimodal therapy, level of education and surgeon factors are the reasons for low uptake of Radical Cystectomy. In addition, marital status and insurance coverage increase the uptake of radical Cystectomy.

## INTRODUCTION

Bladder cancer is the 7<sup>th</sup> most commonly diagnosed cancer in males accounting for 9.0 per 100,000 person/ year (age-standardized incidence rate) globally. In Females, the age-standardized incidence stands at 2.2 per 100,000 person / year. (1)

There is a paucity of data on the prevalence and incidence of muscle-invasive bladder cancer in Kenya. However, data from the county of Nairobi registry in Kenya puts the incidence of bladder cancer at 1.4 per 1,000 000 (age-standardized incidence rate) (2)

Non-muscles invasive bladder cancer (Ta, CIS) accounts for 75%, while 25% is the muscle-invasive bladder cancer (T2 disease) (1)

The gold standard of management of Muscle invasive bladder cancer is radical Cystectomy, but the uptake of this procedure is still low globally. Higher comorbidities and advanced age are among the contributory factors that impact the low uptake of radical Cystectomy for muscle-invasive bladder cancer. (3) Bladder sparing procedure is proposed for patients who do not opt for radical Cystectomy. Trimodal therapy entails Transurethral Resection of the Bladder (Radical) or partial Cystectomy, chemotherapy and radiotherapy. (4)

Despite radical Cystectomy being the gold standard, its uptake or utilization stands as low as 6% and 19-21% for all age groups and patients older than 66 years, respectively. (3) Higher age (greater than 66) correlates with higher comorbidities. Additionally, factors affecting the low uptake of radical Cystectomy include patient, hospital and surgeon (health provider) factors (3)

This study aimed to determine the factors affecting the low uptake of radical Cystectomy at a referral Hospital in Kenya.

## METHOD

### Study procedure

This was a 5-Year retrospective study conducted from 2017 to 2021 at the Kenyatta National Hospital. Sampling was census and data collected included: Patients related data, Hospital data on surgeon volume and surgeon perception on radical Cystectomy.

Information was collected from the records of adult patients with muscle-invasive Bladder cancer (T2-T4aNoMo) who had complete data. Additionally, an online standardized questionnaire was administered to practicing urologists regarding their perception of the factors that affect the low uptake of radical Cystectomy.

A total of 410 patients with Bladder cancer were identified from the records out of which 56 patients had Muscle Invasive Bladder Cancer (T2-T4aNoMo disease). However, only 27 cases met the inclusion criteria. Fourteen practicing urologists responded to the online survey.

Factors assessed included patient factor (age, comorbidities, marital status, education, geographic location, and TNM stage), Hospital factor (Hospital volume), and Urologist perception (Surgeon volume, surgeon skills, surgeon preference for radical Cystectomy, and surgeon for postoperative radical cystectomy quality of life). High Volume Hospital center means a hospital that performs >30 cases/year; Intermediate Volume centers perform 10-29 cases/year) and Low Volume centers <10 cases/Year. Case volume for surgeons under surgeon factor was divided into the following: 2-5 cases/year (lowest case Density), 5-9 cases/year, 10-19 cases/Year, 20-29 cases/year, and ≥ 30 cases/Year. More than 30 cases per year put a surgeon at a high case density and which also impacts the intraoperative and postoperative outcomes. This study also looked at the types of the hospital (Teaching, non-teaching, and county referral hospitals) where the surgeon practiced.

### Data management and analysis

The investigators in this study held a high degree of compliance to research protocol and data quality standards. The principal investigator verified and counterchecked data collected against the selected files to assess the data's accuracy and completeness during the study period. The retrospective data was obtained by pre-trained three (3) research assistants and the principal investigators. A standardized questionnaire was used to capture the data from the file, while a structured online questionnaire was used for the Urologist (surgeon) perception. Data were analyzed by STATA 13.0 and presented in percentages, frequency tables, pie charts, and bar graphs.

### Ethical consideration

Clearance was sought and obtained from the Kenyatta National Hospital Research Department through the Urologic Unit, KNH, to conduct a retrospective study that involved the perusal of medical records of all patients with muscle-invasive bladder cancer. Consent was sort from the Surgeons (Urologists) who partook in the perception survey of factors affecting the low uptake of radical Cystectomy.

### RESULTS

A total of 27 records of patients out of 56 who had muscle-invasive bladder cancer met the inclusion criteria (complete data and T2-T4aNoMo disease). Males accounted for 63%, while females accounted for 37%. Of the 27 eligible patients, 37% had radical Cystectomy, while 55.7 % had Trimodal therapy (Fig 3). The remaining patients (7.4%) declined intervention or were lost to follow-up. Out of the patients who had radical cystectomy only 7.4% were of advanced age (80 years and above). From the data, the youngest patient was 36 years old, and the oldest patient was 94 years (Fig 1). Urban versus rural residence stood at 44.4 and 55.6%, respectively. Of the patients who had Radical Cystectomy, 90 % were married. The majority (62.4%) of the patients who received radical Cystectomy had formal insurance (National Health Insurance Fund), compared to 37% who did not have any form of insurance.

In the radical cystectomy group, 60% had formal education (secondary to college), while 40% had no formal education. In terms of comorbidities, 90% of the patients that got Radical Cystectomy plus urinary diversion had less than 2 comorbidities or no comorbid condition (Fig 2). The ileal conduit was the predominant method of urinary diversion during the study period. In the Tumour Node Metastasis (TNM) staging, 56% had T2NoMo, and 44% had T3NoMo disease.

Under Surgeon factors, 71% had done a radical cystectomy during their practice as urologists. All of the respondents had done 2-5 cases/year of radical Cystectomy plus urinary diversion. Despite the low surgeon case density, 50% were from a Teaching hospital, which is expected to have a high case density. Assessing proficiency, on a scale of 1 to 5, with 5 as the highest proficiency level, 28.6% of the respondents were at level 5 and 7.1% at level 1, with level 3 accounting for 35.7%. All respondents were concerned about the quality of life issues post-operatively and thus influenced their choice of management of Muscle invasive bladder cancer (fig 4).

Data on hospital factors showed that 57.1% of the respondents' facilities had less than five urologists in practice, and 35.7% of the respondents had 5-10 urologists, which points toward low surgeon volume.

Figure 1: Plot Age (age distribution)

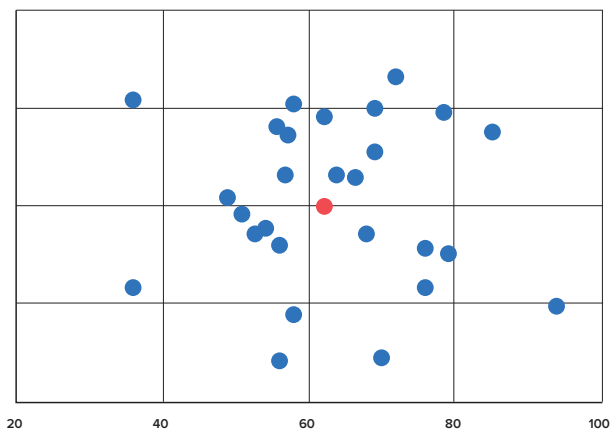


Fig 2 (comorbidities )

Fig 3 (Type of management)

Figure 2: comorbidities

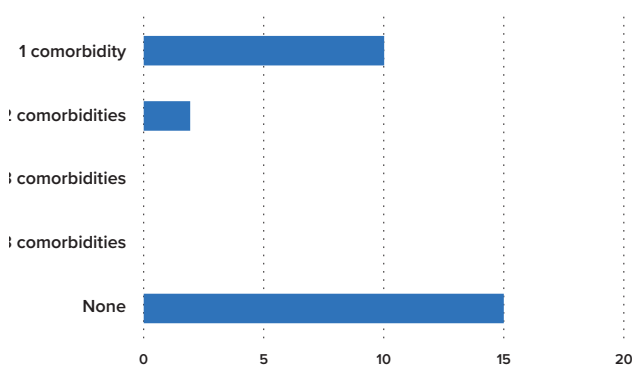


Figure 3: Type of treatment offered.

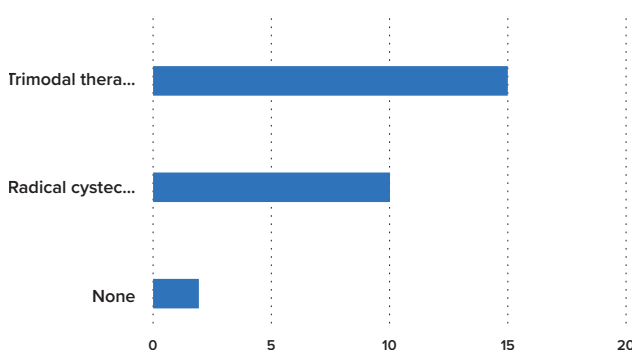
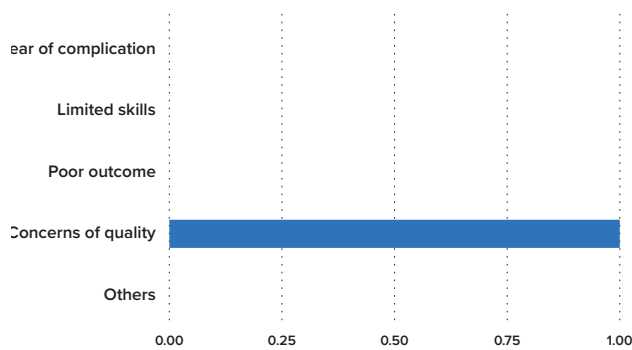


Figure 4: Surgeon perception of Post Op for RC



## DISCUSSION

In this study, 37% of the patients diagnosed with muscle-invasive bladder cancer had radical Cystectomy, but William et al. 2018, put radical cystectomy incidence at 6%. (3) This study found that 55.6% of the patients diagnosed with muscle-invasive bladder cancer had Trimodal therapy instead of Radical Cystectomy. Patients with increased comorbidities and advanced age usually opt for trimodal therapy (3), which is in contrast to the findings in this study where the Trimodal therapy group was younger (66 years and below). Despite the low uptake globally, there is a minimal increase in the uptake of radical Cystectomy in this study compared to the global data. Patients aged 80 years and above who had radical Cystectomy at Kenyatta National Hospital constituted 7.4%, which is slightly congruent with data published by William et al. of a low uptake of between 1% and 10%(3). Of the patients who received Radical Cystectomy, 90% were married, and only 10% did not opt for surgery. Raj S. Pruthi et al. (5) published the Impact of marital status on patients undergoing radical Cystectomy. They reported that 74% of the patients undergoing Radical Cystectomy were married and this is in line with this research finding which is however slightly higher at 90%. Similar observations were made by Canter and colleagues (6) who reported a 71% rate of radical Cystectomy in patients who were married with a p-value <0.001. Majority (62.4%) of the patients who received radical Cystectomy in this study had a formal insurance scheme (National Health Insurance Fund), and 37% did not have any form of insurance or were of unknown insurance statuses. Alexander P. Cole et al. (7) and colleagues concluded that uninsured patients as more likely to die of bladder cancer and less likely to receive neoadjuvant chemotherapy. Patients without insurance coverage were more likely to experience a delay in treatment, particularly radical Cystectomy

In line with the Tumour Node Metastasis (TNM) staging, 70.3% had T2NoMo, and 29.6% had T3a-bNoMo, but no T4 disease was documented during the study period. A greater percentage (70.3%) had localized disease and should have favored a radical cystectomy, but the data showed a higher percentage for trimodal therapy. Localized disease increases the uptake of radical Cystectomy according to Chamie et al. (8). This is in contradiction to the findings of this study in which trimodal therapy was used in the majority of patients who had localized disease.

Various papers support a high rate of radical Cystectomy in teaching institutions (3, 8, 9, 10). Half or 50% of respondents in this study practice urology at teaching hospitals, while the other 50% work in non-teaching hospitals, including county hospitals. All or 100% of the surgeons had a case density of 2-5 cases per Year (low volume) which is different from a report by Chemie et al who found a high case volume and high uptake of radical Cystectomy in teaching hospitals (8). All respondents unanimously agreed on concerns about the postoperative quality of life issues that may negatively influence the uptake of radical Cystectomy. There is debate on the Impact of low surgeon volume on the perioperative outcomes of radical Cystectomy, which then ultimately influence a patient's choice of treatment. Currently, hospital volume (high volume centers- 30 cases/year) favors higher uptake of radical Cystectomy (11) than the surgeon volume. In this study, 57.1% of the respondents had less than five urologists (low surgeon volume) practicing in their facility.

## CONCLUSION

Whereas most guidelines recommend radical Cystectomy for muscle-invasive bladder cancer, there is still a low uptake of this surgical procedure in Kenya. This study found that despite a relatively younger population favoring radical Cystectomy, a significant percentage of patients still opt for Trimodal therapy. Having an insurance cover(NHIF) and marital status are significant factors for increased uptake of radical cystectomy for MIBC among the Kenyan population. The absence of comorbidities, severity of disease, surgeon factors, and level of education does not influence the uptake of radical cystectomy in the same population. There is a need for a prospective study to look at patient perception on the choices of radical Cystectomy versus trimodal therapy/ bladder preserving surgery.

### Limitation

The study being retrospective was impacted by the inadequacy of data and lack of randomization.

### Acknowledgement

Authors' contributions: L.B. Kortimai carried out the literature search, data extraction, statistical analysis, drafted the article, and revised and submitted the article. F. A. Owillah ,J. Ikol ,P. Mburugu, D. Kimani coordinated and participated in the entire process of drafting and revising the article. All authors read and approved the final article.

### Disclosures

No disclosure to make or conflict of interest

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# Laparoscopic Pyelolithotomy - another tool for large renal stones

**Dr. Aideed Kahie<sup>1</sup>, Dr. Abdirashid A Mohamed<sup>1</sup>**

<sup>1</sup> Kaatun Urology Center & Male Executive Clinics

**Corresponding Author:** Dr Aideed Kahie. Email: aideed@kaatun.com

## ABSTRACT

**Background:** Percutaneous nephrolithotomy (PCNL) is the standard treatment for large renal pelvis stones, however, laparoscopic pyelolithotomy (LPL) can be used as an alternative. The aim of this study was to analyze and report our experience with laparoscopic pyelolithotomy in our setting.

**Material and methods:** A retrospective analysis of patient records between 2018-2021 who underwent laparoscopic pyelolithotomy in our setting.

**Results:** The average age was 42 years and the ratio of male to female was 61.5%:38.5% and a mean BMI was 26.1 kg/m<sup>2</sup>. The average stone size was 2.6cm. The stone free rates were 100% at three months. There were no significant complications in the patients.

**Conclusion :** Laparoscopic pyelolithotomy is associated with good perioperative outcomes and is a feasible option for large renal stones in our setting.

## INTRODUCTION

Renal stones are a common benign urological disease that occurs in all age groups(1, 2). Due to the associated morbidity and potential mortality from renal stones, timely, appropriate and effective therapy is necessary(2, 3). There are several treatment modalities for renal stones depending on both stone factors (size, composition, number, and location of the stone) and patient factors (clinical condition of the patient, anatomy of the collecting system, bilateral disease, and obesity) (4, 5). The options of therapy range from medical therapy, non-invasive procedures such as extracorporeal shock wave lithotripsy (ESWL), minimally invasive options like (Percutaneous nephrolithotomy (PCNL), retrograde intrarenal surgery (RIRS), Laparoscopic Pyelolithotomy), and the more invasive options of open pyelolithotomy and anatomic nephrolithotomy(4, 6).

The stone clearance or stone-free rates is an important factor when considering the option of treatment(7). It is dependent on stone bulk or burden, location, composition, and anatomy of the collecting system. It is preferable if all the stones can be cleared in one session, but at times due to the complexity of stone distribution more than one session is required to clear all the stones completely which is associated with increased cost and morbidity(6, 7).

Most guidelines recommend that large calculi (>2cm) or complex renal calculi should be managed with percutaneous nephrolithotomy (PCNL) because of the increased morbidity associated with open surgery. PCNL has a high stone clearance but is associated with increased morbidity such as renal parenchymal injury, significant hemorrhage and sepsis(7, 8). While there are reports of miniaturized PCNL techniques which are associated

with decreased blood loss, they are also associated with significantly longer operation time. Retrograde intrarenal surgery (RIRS) is another alternative for treating renal stones by stone fragmentation and dusting under endoscopic direct vision. RIRS is associated with lower morbidity compared to PCNL but its disadvantage is prolonged procedure time for larger stones, poorer stone clearance and hence often the need for a second procedure(9).

As laparoscopic techniques have been improving there has been an increased use of laparoscopic pyelolithotomy (LPL) as a modality of treatment for large and complex renal stones. The advantage of LPL is the whole stone can be retrieved intact thus a higher stone free rates, good visualization, reduced pain, bleeding and morbidity that is seen with PCNL(7). Laparoscopic pyelolithotomy is indicated in cases with complex stone burden, complex renal anatomy, failed endoscopic procedures, concomitant surgery such as pyeloplasty, and contraindication for PCNL (e.g. retro-renal colon)(5, 6).

Recently several studies have compared LPL vs PCNL in management of large renal stones. 2 metanalysis noted LPL was associated with higher stone free rates and lower morbidity and retreatment rates. However, LPL had a longer operating time(7, 9).

There is a paucity of data on the use of LPL in the treatment of large renal stones in our region. The aim of our study is to report our experience in using laparoscopic pyelolithotomy in management of large and complex renal stones.

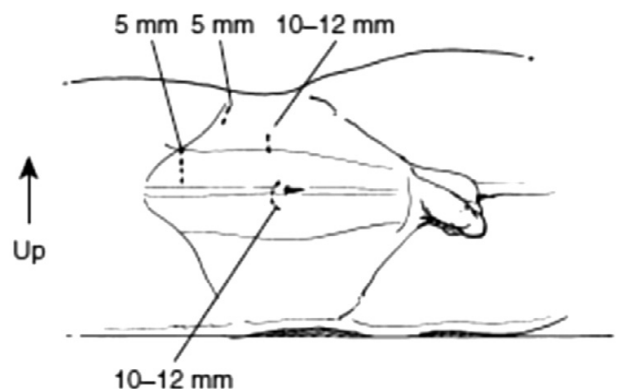
## METHODOLOGY

This was a retrospective study of all the patients who underwent Laparoscopic pyelolithotomy between 2018-2021. Data collected from patients' medical records included demographic and clinical characteristics. Radiological information collected including stone size, location and anatomical anomalies. Surgical outcomes were assessed as site of surgery, operative time, estimated blood loss (EBL), postoperative complications and length of hospital stay. IBM Statistical Package for Social Sciences (SPSS) Version 25.0 (IBM Corp, Armonk, NY) was used for statistical analysis. Descriptive characteristics were summarized and presented as means and standard deviation. Categorical variables were presented as frequencies and proportions

## Technique of laparoscopic pyelolithotomy

Patients were placed in a modified lateral decubitus position with minimal flexion. Paraumbilical visual optical port is inserted and after pneumoperitoneum is achieved, three or four working ports are inserted under vision. (Fig. 1) LP was done via transperitoneal approach with medial reflection of colon to access retroperitoneum. Liver and spleen are retracted superiorly for safe exposure of the renal hilum. The ureter is identified over the psoas muscle and is traced up to the renal pelvis.

Figure 1: Port location for laparoscopic transperitoneal approach



Normally the pelvic stone is identified by its bulge. If identification is difficult due to inflammation or adhesions, then exploration of the renal hilum is done and the renal vessels are identified and protected. Pyelotomy is done as in open surgery in a v-shaped manner with creation of a flap using laparoscopic scissors/ via transverse fashion. (fig.2) Stone in the pelvis are retrieved with stone grasping forceps. The entire pelvicalyceal system is evaluated using flexible cystoscope and calyceal stones are retrieved using grasper or basket. (Fig. 3, 4) Retrieved stones are placed in a handmade rubber glove basket under vision. (Fig. 5) A 6/24 F Double-J stent is passed over a guidewire via pyelotomy incision with the distal end in the bladder and the proximal coil reinserted into the renal pelvis after the guidewire is removed. The incision is closed using 4/0 polydioxanone (PDS) sutures. Occasionally a JP drain is placed in the pararenal space. The stones are extracted via inferior most port and if necessary, the incision is extended. All port sites are closed with absorbable monofilament suture.



Figure 2: **Pyelotomy done with hook electrocautery and retrieve calyceal stones**

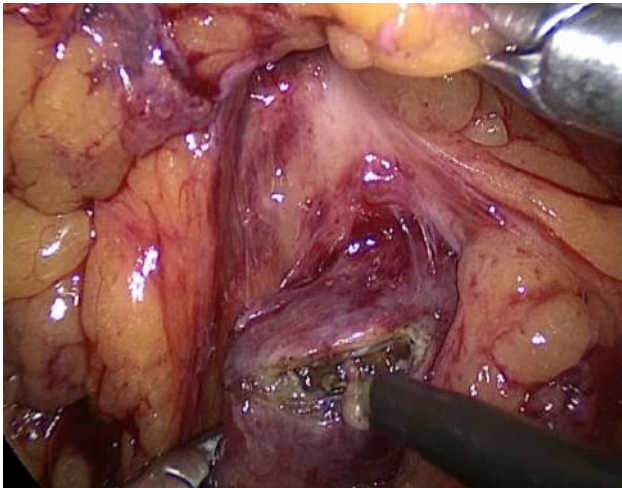


Figure 3: **Using a flexible scope to evaluate the calyces**

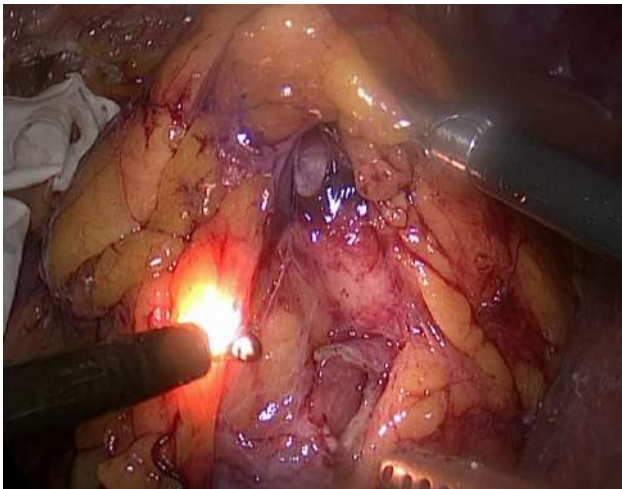


Figure 4: **Stone being retrieved using basket**

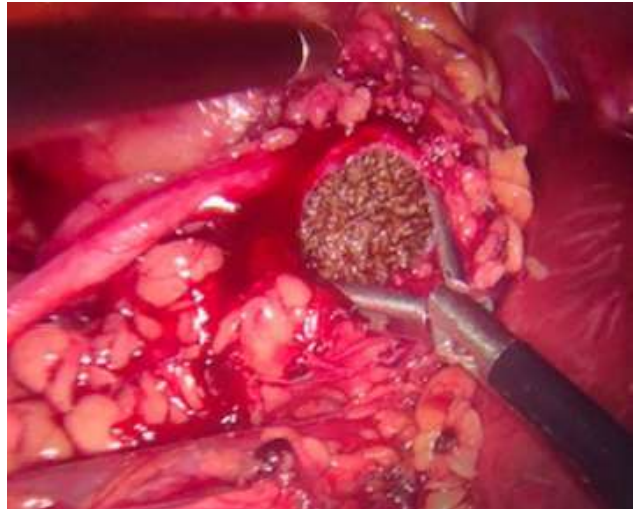


Figure 5: **Placement of retrieved stone in a glove bag**



## RESULTS

Thirteen cases were recruited into the study. The minimum age was 16 and a maximum was 74 yrs. The male to female ratio was 61.5% vs 38.5%. The mean BMI was 26.1 kg/m<sup>2</sup>. One patient (7.6%) had diabetes mellitus and 2 patients (15.4%) had hypertension. The mean stone size was 2.6 cm. The stones were all located in the renal pelvis and no anatomical anomalies were noted as shown in Table 1.

The mean operative time was 137.7 ± 25 and there was no significant blood loss reported (30- 120mls) Postoperative course was unremarkable in our patients only one patient developed fever and wound (port) site infection. The average length of stay for our patients were 2.3 ± 0.72 days.

Table 1: Demographic Profile, Sone Characteristics

Patient characteristics	
Mean age (years)	42.7± 13.2 (16-71)
Gender Male	8 (61.5%)
Female	5 (38.5%)
Mean BMI kg/m <sup>2</sup>	26.1 ± 4.04 (19-32)
Comorbidities n%	
Diabetes Mellitus	1 (7.6%)
Hypertension	2 (15.4%)
Mean stone size, cm	2.6 ± 0.42 (2.2-3.8)

Table 2: Surgical Outcomes

Stone free rates no. (%)	13 (100%)
Mean operation time (min)	137.7 ± 25
Mean estimated blood loss (ml)	54.62±19 (30-100)
Mean length of hospital stay (days)	2.3 ± 0.72
Fever	1 (7.7%)
Wound site infection	1(7.7%)
Port site hernias	0

## DISCUSSIONS

Minimally invasive surgery in urological practice has improved significantly since the first laparoscopic nephrectomy by Clayman et al in 1991(10). Laparoscopy has many advantages as it associated with less postoperative pain, decreased morbidity and reduced length of hospital stay. The uptake of minimally invasive surgery in urology in our region has been slow due to the unavailability of the equipment and the technical expertise(11, 12) . Laparoscopic pyelolithotomy is considered a second line to PCNL for management of large renal stones >2cm as it is considered to be more invasive and less cosmetic compared to PCNL. The aim of this study was to demonstrate the outcomes of LP in our setting. Thirteen LPLs were performed by a single laparoscopic urologist in tertiary institutions in the city.

The demographic and stone characteristics of our patients were comparable to other studies. The mean operative time was 137.7 ± 25 which is similar to the study by Meria et al and Al Hunayan et al (5, 13). They noted the operative time was longer compared to PCNL and attributed it to the learning curve of LPL and time required for intracorporeal suturing and delivery of the stone into the endobag. None of the LPLs were converted to open in our study. The average estimated blood loss in our study was 55 mls and

none of our patients required transfusion which is similar to reports in the literature(4). A meta-analysis by Bai et al noted PCNL patients had higher bleeding rates (OR 0.20, 95% CI 0.06–0.61, P = 0.005) and transfusion requirement (OR 0.28, 95% CI 0.13–0.61, P = 0.001) compared to LPL(7).

The complications noted in our study were one patient developed fever due to wound site infection which led to a longer inpatient period. This has been reported in other studies that noted a similar number of patients developed fevers which prolonged their hospital stay(14). PCNL is associated with higher incidences of fever compared to LPL which most of the time resolves but a few cases are usually associated with urosepsis (7, 15, 16). The mean length of hospital stay (LOS) for our patients was 2.3 days which was similar to other reports in literature

The stone free rates in our study were 100% after three months review. This is similar to reports by Al Hunayan et al and was mainly attributed to intact removal of the stone, unlike PCNL where the stone is fragmented which may leave some residual fragments(13, 14). Studies comparing PCNL and LPL noted LPL was associated with higher stone free rates although not statistically significant (7, 13, 14). There were no long-term complications or mortalities noted in our study.

Although PCNL is the gold standard for large complex renal stones, traversing the renal parenchyma can be associated with significant complications such as bleeding, nephron damage, urine leakage and urosepsis (7, 9, 14). Even though LPL is more invasive than PCNL, as it requires 3-4 trocars to be placed compared to a single puncture in the latter, it is better when compared to morbidity of open surgery. There are studies which have reported cases of solitary renal pelvis stone which were ideal for ESWL and PCNL, but as these modalities were not available and LPL was a feasible option for these patients thus avoided morbidity of open surgery. (14, 17) Other studies have also recommended LPL use in cases of large stone burden in multiple locations in the kidney(4, 6, 18).

This study has demonstrated LPL is a feasible option in management of complex renal stones especially in cases where PCNL and ESWL are not available or in a complex hostile renal anatomy. Like other minimally invasive techniques LPL has a learning curve in which trainees improve and reduce complication rates. The relatively small heterogenous sample size is an important limitation of the study.

## CONCLUSION

PCNL is the standard management for large complex renal stones and LPL has been demonstrated to be an alternative especially in areas where PCNL is not available. LPL is associated with a good favorable outcome and is feasible in our setting.

## Authors contribution

Both authors contributed to the design of the study as well as data analysis (Dr. Aideed Kahie, Dr. Abdirashid A Mohamed). The manuscript's author is Dr. Aideed K. After being critically edited by both writers, the text's final version has been approved for publishing.

## Conflict of interest

The authors declare no conflict of interest

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# BCG therapy for high-risk NMIBC: A national referral hospital experience

**Ernest N Kimani<sup>1</sup>, James A. Iko<sup>2</sup>, Patrick G. Mburugu<sup>2</sup>, Francis A. Owilla<sup>1</sup>**

<sup>1</sup>Department of Surgery, University of Nairobi.

<sup>2</sup>Department of Surgery, Kenyatta National Hospital.

**Corresponding Author:** Dr Ernest Kimani. Email: drenk10@gmail.com

## ABSTRACT

**Introduction:** The treatment of choice for high-risk non-muscle invasive bladder cancer (NMIBC) is bacillus Calmette-Guérin (BCG) therapy. However, BCG accessibility and use in high-risk bladder cancer patients have been full of significant challenges including shortages, cost concerns, and unfavorable side effects profile limiting its use. This has a risk of increasing bladder cancer recurrence and progression rates, especially in low- and middle-income areas of the world.

**Objective:** The aim of this study was to investigate the uptake of BCG therapy for high-risk bladder cancer in patients seen at a national referral hospital in Kenya

**Methodology:** Through a retrospective cohort study design and consecutive sampling approach, patients who were diagnosed with high-risk NMIBC between January 2018 – December 2021 were recruited, and their past medical records were assessed on bladder cancer grade and stage, and whether they received BCG therapy. Dependent variables were the use of BCG, recurrence and progression rates of bladder cancer, and independent variables were age, sex, and number of BCG sessions administered. Urologists in the department were similarly interrogated on the barriers to utilization of BCG if any. Stata 16 was used for data analysis.

**Results:** Twenty-six patients were diagnosed with high-risk bladder cancer. The mean age was 62 years, (SD 14.67, Range 29 – 88). Males were 14 (53.9%), with females being 12 (46.2%). Ta tumors were 8 (30.8%) and T1 tumors were 19 (69.2%). According to stage and grade, the Ta-low grade was 15.4%, the Ta-high grade was 19.2%, the T1-low grade was 15.4%, and the T1-high grade was 50%. The uptake of BCG was only in 2 (7.7% [CI 0.9 – 24.3]) cases. Of the two patients, none received BCG therapy according to the expected timelines as defined by the guidelines. Of the two patients, one received two cycles and one received only one induction cycle. At the end of the study period, the recurrence rate was 64.7% (CI 38.3 – 85.8%) and a median recurrence time of 12 months. The Progression rate for bladder cancers was 35% (CI 14.2 – 65.7) with a median time to progression of 24 months. Barriers to uptake of BCG therapy were reported as shortages, logistics, storage and handling difficulties, cost concerns, inappropriate follow-up, of patients, and unfavorable side effect profiles.

**Conclusion:** High rates of bladder cancer recurrence and progression are directly correlated with poor rates of BCG uptake among high-risk NMIBC. The low BCG installation rates are caused by significant obstacles, and novel strategies are needed to improve uptake. This would in turn lower morbidity, mortality, and costs of managing bladder cancer.

**Keywords:** BCG utilization, Bladder cancer, High risk, Recurrence, Progression

## INTRODUCTION

Bladder cancer is the ninth leading cause of death in developed countries, and the second most common urological malignancy. There were 2.63 million Bladder Cancer cases estimated from the GBD 2017 data, with 200,000 persons dying of Bladder Cancer, resulting in 3.60 million DALYs in 2017. The age-standardized prevalence (ASP) of Bladder Cancer was 32.91/100,000 persons, and the age-standardized death rate (ASDR) was 2.57/100,000 persons (1).

The 8<sup>th</sup> edition of the American Joint Committee on Cancer (AJCC) staging system is adopted for the staging of bladder cancer. Ta is a tumor localized to the epithelium. T1 are tumors invading the lamina propria. T2 tumours invade the detrusor muscle with T2a denoting superficial muscle invasion and T2b denoting deep muscle invasion. T3 tumours are now defined as those that extend beyond the bladder to involve the perivesical fat, either microscopically (T3a) or macroscopically (T3b). T4 Tumors extend to the pelvic organs with T4a involving the prostate, uterus or vagina and T4b involving the abdominal or pelvic wall (2). Tumors confined to the epithelium and lamina propria are termed as non-muscle invasive bladder cancers (NMIBC) and represent 70 – 75 % of bladder cancers while those invading the detrusor muscle are termed as Muscle invasive bladder cancers (MIBC) and comprise 20 – 25%. Metastatic tumors comprise about 5 – 10% (3).

NMIBC can be classified as low risk, intermediate risk and high risk (4). The European Organization for Research and Treatment of Cancer (EORTC) defines high risk bladder cancers as non-muscle invasive tumors with the following characteristics, T1, high grade, carcinoma in situ and Ta large (>3cm), multiple and recurrent (5). However, variations amongst guidelines exist in defining the risk stratification of non-muscle invasive tumors (4,5).

The standard recommended and most effective treatment of high risk NMIBC is the intravesical administration of Bacillus Calmette-Guerin (BCG) (6). BCG induces a non-specific promotion of the biological function in cells of the innate immune system, thus creating a heterologous immunological memory that has been termed trained immunity. The result is stimulation of innate immune cells to mount a more robust response to secondary non-related stimuli after being initially primed.

This effect contributes to the remission of the bladder cancer (7) the mechanism of its therapeutic effect is still under investigation. The requirements for effective BCG therapy include an intact immune system, live BCG, and close contact of BCG with bladder cancer cells. Important constituents of the cellular inflammatory response to BCG include CD4+ and CD8+ lymphocytes, natural killer cells, and granulocytes. Important elements of the humoral immune response to BCG include TRAIL (tumour necrosis factor-related apoptosis-inducing ligand). However, the exact mechanism of BCG in controlling bladder cancer are complex.

High risk bladder cancer has high recurrence and progression rates. One study estimated a 10-year recurrence of 74.3%, progression of 33.3% and mortality of 12.3% in 7410 patients with high risk bladder cancer (8) progression and bladder cancer-related mortality rates in a cohort of individuals with high-grade non-muscle-invasive bladder cancer.

**Methods**

Using linked SEER-Medicare data, we identified subjects with a diagnosis of high-grade, non-muscle-invasive disease in 1992–2002 and were followed until 2007. We then used multivariate competing-risks regression analyses to examine recurrence, progression, and bladder cancer-related mortality rates.

**Results**

Of 7,410 subjects, 2,897 (39.1%). BCG use in high-risk bladder cancer is highly effective and has been shown to result in upto 80% complete response (9,10) T1, Tis. However, BCG accessibility and use in high risk bladder cancer patients has been fraught with significant challenges including shortages, cost concerns and unfavorable side effects profile limiting its use (6). This has a risk of increasing bladder cancer recurrence and progression rates. Thus the main aim of this study was to investigate the rates of utilization of BCG, associated recurrence and progression rates and barriers to BCG utilization at a national referral hospital.

## MATERIALS AND METHODS

### Study design, site, period, population, inclusion and exclusion criteria.

A retrospective cohort study of patients who underwent treatment for high-risk bladder cancer at a national referral hospital between 2018 – 2021 was conducted. Consecutive sampling approach was used. High risk bladder cancer was defined according to the EORTC definition.

Inclusion criteria was all patients diagnosed with high-risk bladder cancer and eligible for administration of BCG. Outcome data was collected on rates of utilization of BCG, recurrence and progression rates of bladder cancer. Exposure data was collected on demographics and tumor characteristics.

Urological surgeons in the facility were similarly interrogated on the perceived challenges facing utilization of BCG.

**Data analysis**

Stata 16.0 was used for data analysis. Descriptive statistics (frequencies, proportions, means and standard deviations) were used to determine the socio-demographic and clinical characteristics of the study participants. To calculate the rate of utilization of BCG therapy in high-risk bladder cancer, number of patients who were prescribed for and received intravesical BCG therapy for high-risk bladder cancer was divided by the total number of patients eligible over the study period. To assess time to recurrence of bladder cancer, Kaplan Meir survival curves were used. Results of the survival model were reported in Hazards ratios and corresponding 95% confidence intervals and p values where significance level shall be <0.05.

**RESULTS**

There were 26 patients diagnosed with high-risk bladder cancer. Mean age was 62 years, SD 14.67, Median 62, Range 29 – 88. Males were 14 (53.9%); Females were 12 (46.2%).

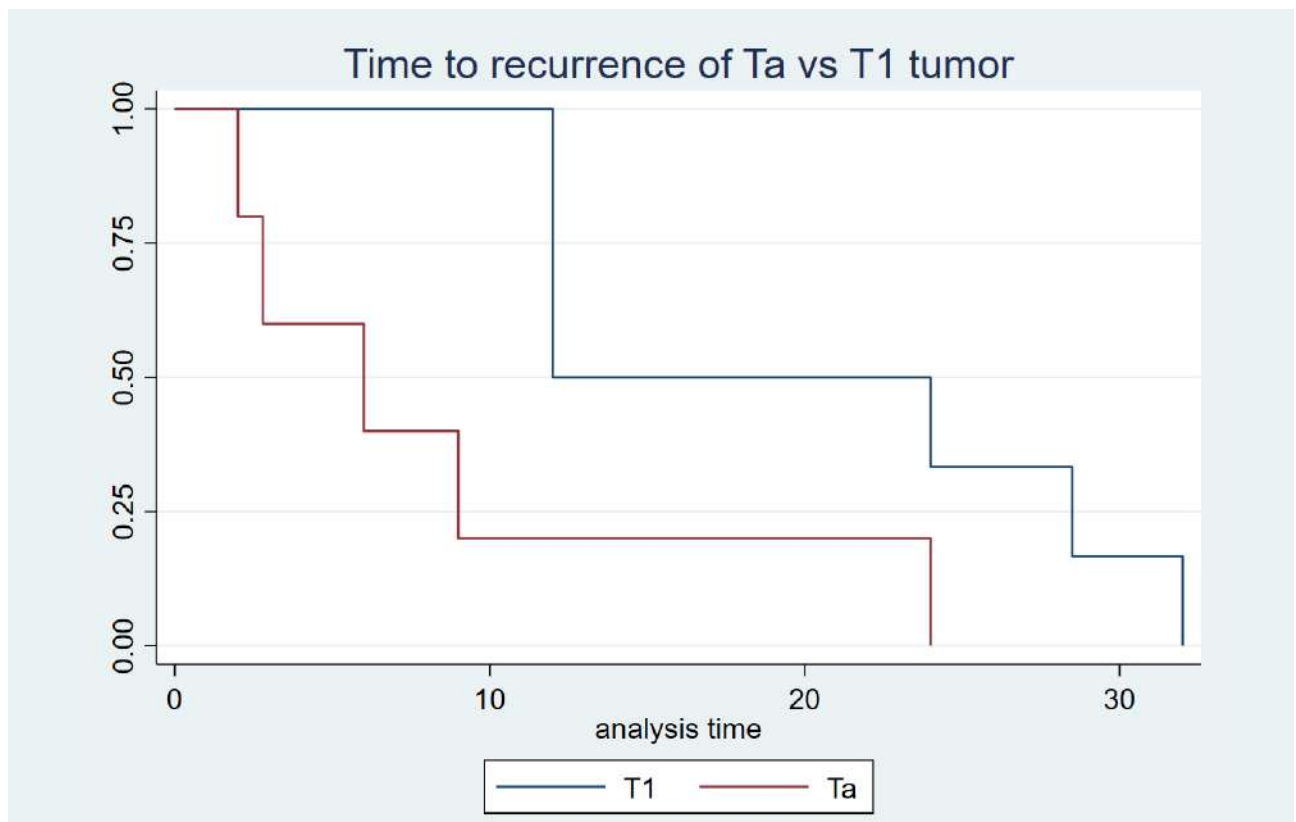
According to the stage of tumors, Ta tumors were 8 (30.8%) and T1 tumors were 19 (69.2%). According to stage and grade, Ta-low grade were 15.4%, Ta-high grade (19.2%), T1-low grade 15.4% and T1-high grade 50%.

The uptake of BCG was only in 2 (7.7% [CI 0.9 – 24.3]). Of the two patients, none received according to the expected timelines as defined by the guidelines. Of the two patients, one received two cycles and one received only one induction cycle.

At the end of the study period, the recurrence rate was 64.7% (CI 38.3 – 85.8%) and a median recurrence time of 12 months.

Time to recurrence in comparison of Ta vs T1 tumors demonstrated that the Ta tumors had a greater hazard of recurrence, Hazard ratio of 4.64 (95% CI 1.2 – 18.2), p value 0.028 (Figure 1).

Figure 1: Kaplan Meir curve of Time to recurrence of Ta vs T1 tumor.



Progression rate for the bladder cancers were 35% (CI 14.2 – 65.7) with a median time to progression of 24 months.

In a survey of 12 urology consultants and residents at the department, 83% noted that there were significant barriers hindering patients to access or receive the medications. The barriers are listed in Table 1.

Table 1: **Barriers to BCG therapy in high-risk bladder cancers.**

Challenge	Frequency
Unavailability due to shortages	10 (83.3%)
Inability to afford by patients	8 (66.7%)
Logistics difficulties (transport and storage)	8 (66.7%)
Lack of clear follow-up of patient on treatment protocols	5 (41.7%)
Unfavorable side-effect profile	2 (16.7%)
Others	1 (8.3%)

## DISCUSSION

High risk bladder cancer is increasingly common. The mean age of patients in our study was 62 years with males being 53.7% and females 46.2%. A systematic review conducted by Musat et al, 2021, on high risk bladder cancer patients revealed a mean age of 71 (65 - 86) years in high risk bladder cancer where males were 79% (56 – 100%) and females, 21% (11). Studies demonstrate that bladder cancer is generally four-fold higher in males than females (12). Our study further demonstrated that Ta tumors were 8 (30.8%) and T1 tumors were 19 (69.2%). In accordance to stage and grade, Ta-low grade were 15.4%, Ta-high grade (19.2%), T1-low grade, 15.4% and T1-high grade, 50%. Studies show that of all NMIBC, 70% are Ta, pT1 are 20% and CIS are 10% (13)recurrence, and progression of NMIBC and the state-of-the art treatment for this disease.

Evidence acquisition

A literature search in English was performed using PubMed and the guidelines of the European Association of Urology and the American Urological Association. Relevant papers on epidemiology, recurrence, progression, and management of NMIBC were selected. Special attention was given to fluorescent cystoscopy, the new World Health Organisation 2004 classification system for grade, and the role of substaging of T1 NMIBC.

Evidence synthesis

In NMIBC, approximately 70% of patients present as pTa, 20% as pT1, and 10% with carcinoma in situ (CIS). Further studies would be needed to elaborate on epidemiology of high-risk bladder cancers.

The uptake of BCG was only 2 (7.7%). In the systematic review by Musat et al, 2021, 43 (70%) studies reported a BCG instillation rate of 100% whereas 18 studies demonstrated BCG instillation rates of 3 – 86%. This

demonstrates variation in BCG utilization across different environments. Further report indicated that out of 35 studies, 27 (77%) used induction phase in all patients while 8 studies reported an induction therapy rate of 29 – 95% (11).

Global shortages of BCG have been widely reported (6,14). The effect of this shortage globally, was limitation of access of BCG to only patients with high grade disease and carcinoma in situ. Patients were similarly denied maintenance therapies and urologist resulted to using lower dosages (14). In our case low BCG instillation rates are the key outcomes of the BCG shortages. Cost concerns, logistic and handling difficulties, side effect profile and lack of adequate follow-up with patients are reported in our study as contributing to low BCG utilization rate.

BCG therapy has direct influence on recurrence and progression of bladder cancers. In this study, the recurrence rate of the high-risk bladder cancers was 64.7% and a progression of 35%. Of the two patients who received BCG therapy, none showed recurrence and/or progression at the end of the study. Data demonstrates a 5-year recurrence free survival of 17 – 89%. Progression free survival is reported to be 58 – 89%. Cancer specific survival is 71 – 96% and an overall survival is 28 – 90%. The greatest benefit is seen in patients undergoing BCG instillations (11). Patients treated with BCG have as much as 40% reduction in tumor recurrence and is demonstrated to eradicate the tumor in over 60% for papillary disease and 70% for carcinoma-in-situ (CIS)(10). Other studies have demonstrated a recurrence free survival of 73% and 46% at 2 and 5 years respectively after BCG therapy and a progression free survival rate of 89% at 56 months in cases where only induction phase of BCG is given (9)T1, Tis.

## CONCLUSION

BCG instillation is generally inadequate according to the findings of this study. Similarly, recurrence and progression rates of high-risk bladder are generally high. Thus, measures to improve accessibility and utilization of BCG would be paramount to lower the recurrence and progression rates of bladder cancer. Further studies are needed to elaborate on epidemiology of high-risk bladder cancer to understand its impact on morbidity and mortality in patients with bladder cancer.

## LIMITATION

This study was limited by small sample size and retrospective nature of the study hence missing records.

## ACKNOWLEDGMENTS

E. N Kimani, J. A. Ikol, P.G. Mburugu, and F. A. Owilla planned and wrote the manuscript; E. N Kimani critically revised the manuscript; all authors edited and approved the final version

## CONFLICT OF INTEREST

None

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## Presence of Paraganglioma in a Special population- Pregnant Women

**Dr SNK Waweru**<sup>1</sup>- Consultant Urologist, Mater Hospital

**Dr Sophy Opiyo**<sup>1</sup>-The Mater Misericordiae Hospital, COSECSA year III surgical resident.

**Dr Macharia Kanjumba**<sup>1</sup>-The Mater Misericordiae Hospital, COSECSA year V surgical resident

<sup>1</sup>The Mater Misericordiae Hospital

**Corresponding Author:** Dr SNK Waweru. Email: skabakiwaweru@gmail.com

### ABSTRACT

**Background:** Paragangliomas are also known as Extra adrenal Phaeochromocytoma, and their incidences occur in about 10% of the general population. In the pregnant population, studies show an incidence rate of <0.005%. Presentation of a Paraganglioma in a pregnant woman is like that of a non-pregnant woman therefore its management in a pregnant woman is of key importance as delayed management or no management would lead to increased morbidity and mortality to both the neonate and the mother. This case report explains the approach to and subsequent management of a pregnant woman with a paraganglioma.

**Methods:** A case report. A rare case of paraganglioma in a pregnant woman in her second trimester.

**Results:** 28yr old female, para 1+1 on her 3rd pregnancy, presented from the antenatal clinic with persistent high blood pressure and controlled blood sugar levels despite being on anti-hypertensives. She also lost her second pregnancy due to elevated blood pressure- preeclampsia and the initial delivery was via cesarean still due to elevated blood pressure. Her complaints were excessive sweating, increased heart rate and anxiety with a feeling of impending doom. Reported perceiving normal fetal movement. Following thorough evaluation found to have markedly elevated normetanephrine levels and imaging revealed a left extra adrenal mass. Patient was diagnosed with a paraganglioma, started on alpha adrenergic receptor blockage (Phenoxybenzamine) and scheduled for elective excision of the mass, which was done successfully. Post procedure patient reported resolution of her symptomatology, and delivered a live male infant at 34 weeks, with both mother and child doing exceptionally well.

**Conclusion:** Paragangliomas in pregnancy are a very rare occurrence, but clinicians should have a high index of suspicion for those patients who come with a history of high blood pressures, pregnancy loss. Management for such patients is early identification, initialization of alpha-adrenergic receptor blockage, excision of the mass/es during the 2<sup>nd</sup> trimester, and continued monitoring of mother and fetus to achieve a term pregnancy via cesarian section. With such an approach, morbidity and mortality for mother and fetus are markedly reduced.

**Keywords:** Paraganglioma, Phenoxybenzamine

## INTRODUCTION

Paragangliomas are extra-adrenal chromaffin cell tumors, which, in a recent review of their incidence in pregnancy, were reported to comprise only 19% of chromaffin cell tumors, whereas the majority were pheochromocytomas (5). The clinical and biochemical patterns of pheochromocytomas may differ when compared with paragangliomas. Differences in tumor size, catecholamine content, and secretion may result in differential hemodynamic effects. As a result, the maternal and fetal risk may vary according to the tumor site (extra-adrenal).

Pregnancies complicated by a paraganglioma are very rare, estimated to occur in 0.007% of all pregnancies (1). Due to the rarity of paragangliomas in pregnancy, the recommended management for the mother and baby is based on case reports, small case series, and expert opinion.

Improving maternal and fetal mortality in patients with paragangliomas in pregnancy have been published since the initial case reports of the 1950s. In the 1960s, maternal and fetal mortality in paragangliomas was 55% (2). In the period between 1980 and 1987, maternal and fetal mortality in this condition was cited as 26% (3). These respective mortalities in pregnancy-associated paragangliomas decreased further to 17% in cases reported from 1998 through 2008 (4). The diagnosis of paragangliomas is increasingly being recognized in the antenatal period, resulting in 12% fetal mortality (and 0% maternal mortality), compared with 29% fetal and maternal mortality when the diagnosis is made during labor or immediately postpartum (5).

Current recommendations for the management of a paraganglioma discovered in pregnancy are that if the condition is diagnosed in the first 24 weeks of gestation, the tumor should be removed, preferably by laparoscopic approach in the second trimester (6), whereas when a paraganglioma is discovered in the third trimester, surgical excision should be delayed until the fetus is viable and able to be delivered, with the tumor being removed either immediately after delivery or at a later date (6). Due to historical data, in which fetal mortality from vaginal delivery was much higher, cesarean section is the recommended mode of delivery of the baby (7). For imaging, MRI is recommended for paragangliomas in pregnancy due to the associated risk of fetal exposure to ionizing radiation with computed tomography imaging. Preoperative  $\alpha$ -receptor

blockade should be given as for nonpregnant patients (although a target blood pressure (BP) has not been established) along with increased salt and fluid intake (6). Phenoxybenzamine does cross the placenta and may cause neonatal hypotension and respiratory depression (8).

We report our experience of managing paraganglioma in pregnancy when detected early and utilizing guidelines developed from those who have dealt with this rare condition in the past.

## METHOD

Our patient was a 28yr old female, para 1+1 on her 3rd pregnancy, who presented from the antenatal clinic at 21 weeks gestation with persistent high blood pressure and controlled blood sugar levels despite being on anti-hypertensives (Methyldopa and Labetalol). The patient was being treated as having gestational hypertension. She had a past obstetric history of losing her second pregnancy due to elevated blood pressure- preeclampsia. Her index pregnancy was delivered via cesarean section due to elevated blood pressure. Her complaints at this time were excessive sweating, increased heart rate, and anxiety with a feeling of impending doom. Reported perceiving normal fetal movement

Clinical examination noted a short stout woman who was obviously diaphoretic but stable.

Vital signs taken included a blood pressure of 151/100mmHg and a repeat giving 162/94mmHg, pulse rate of 100 bpm, temperature of 36.2, SPO2 of 97% in room air, and random blood sugar of 11.5g/dl

Abdomen examination revealed normal fullness with a fundal height of approximately 22wks, non tender abdomen with bowel sounds present. In the respiratory exam she had bilateral vesicular breath sounds.

The rest of her examination was normal

Laboratory Tests and imaging studies were then conducted. They included a complete blood count, Kidney function test, Thyroid function test, Serum Parathyroid and Cortisol levels, urine Catecholamines and metabolite evaluation. Imaging studies conducted on her included an ECG, Transthoracic echocardiogram, and MRI of the abdomen.

Her results are tabulated in Table 1 and Figures 1 and 2 below

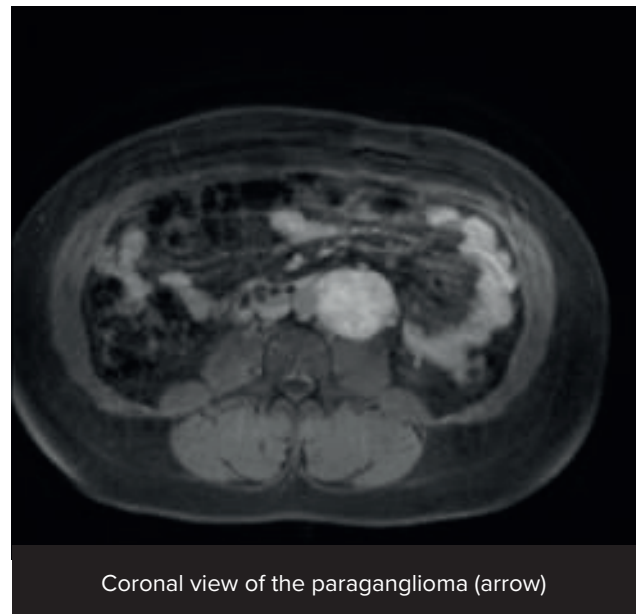
Table 1: Tests done

TESTS		Results
Laboratory	Full haemogram	WBC 11.5, Haemoglobin 11.7, platelets 432
	Urea, creatinine and electrolytes	Na 136, K+ 3.4, urea 3, Crea 60
	Thyroid function tests	TSH 2.67( 0.35- 4.94), FT4 11.41( 9.01- 19.05), FT3 3.56( 2.63- 5.70)
	Serum Parathyroid	36.4 (15-65)
	Serum Cortisol	260nmol/L
	Urine catecholamines and metabolites	Urine pH- 1.0 U- volume- 1330 Urine Creatinine- 12.4(5.3- 15.9) 24hr normetanephrine <b>36450</b> Normetanephrine: creatinine ratio: <b>2934.26</b> 24hr Metanephrine excretion 479(152- 913) Metanephrine: creatine ration 38.54
Imaging	ECG	Normal sinus rhythm
	Transthoracic Echocardiogram	Mild concentric left ventricular hypertrophy, LVEF 55%
	MRI ABDOMEN	4.8 X 4.1 X 4.5 Left extra adrenal paraganglioma

Figure 1: Saggital view of abdominal MRI



Figure 2: Coronal view of abdominal MRI



From the imaging results and laboratory evaluation, a diagnosis of paraganglioma was made. The Patient was appraised on the results of her tests and made to understand that the tumor is the most likely cause of her persistent high pressures and symptomatology, as well as the fetal demise of her second pregnancy. The Patient was then taken through the management of her condition which she understood and accepted. She was started on an alpha-adrenergic receptor blocker- Phenoxybenzamine 10mg once a day for 2 weeks before admission for elective surgery. She reported a reduction in the amount of sweating she had been experiencing during the 2 weeks of treatment and minimal reduction in the blood pressure levels as charted in a book.

She continued using the Phenoxybenzamine till the morning of the procedure with an addition of Nebivolol a beta blocker.

Preoperative management included Phenoxybenzamine 10mg + nebivolol 5mg OD and propranolol 40mg BD, intravenous fluids (Normal Saline), increased oral salt intake, and optimization of blood glucose levels.

Intraoperatively anesthetic management included Glucose-insulin potassium infusion- (1/2 liter of dextrosaline+ 10mg potassium chloride+ 8IU insulin), 2 liters of Ringers lactate, and infusions of nitroglycerin 50ug/hr, noradrenaline 40ug/ml.

Figure 3: Intraoperative view of the mass

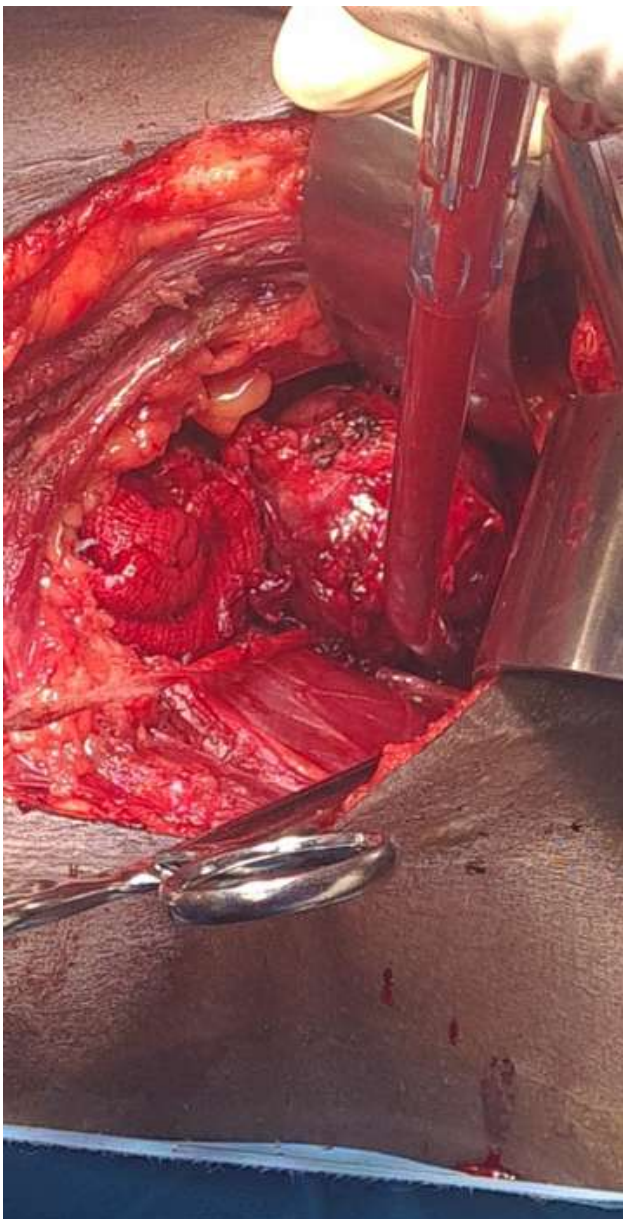
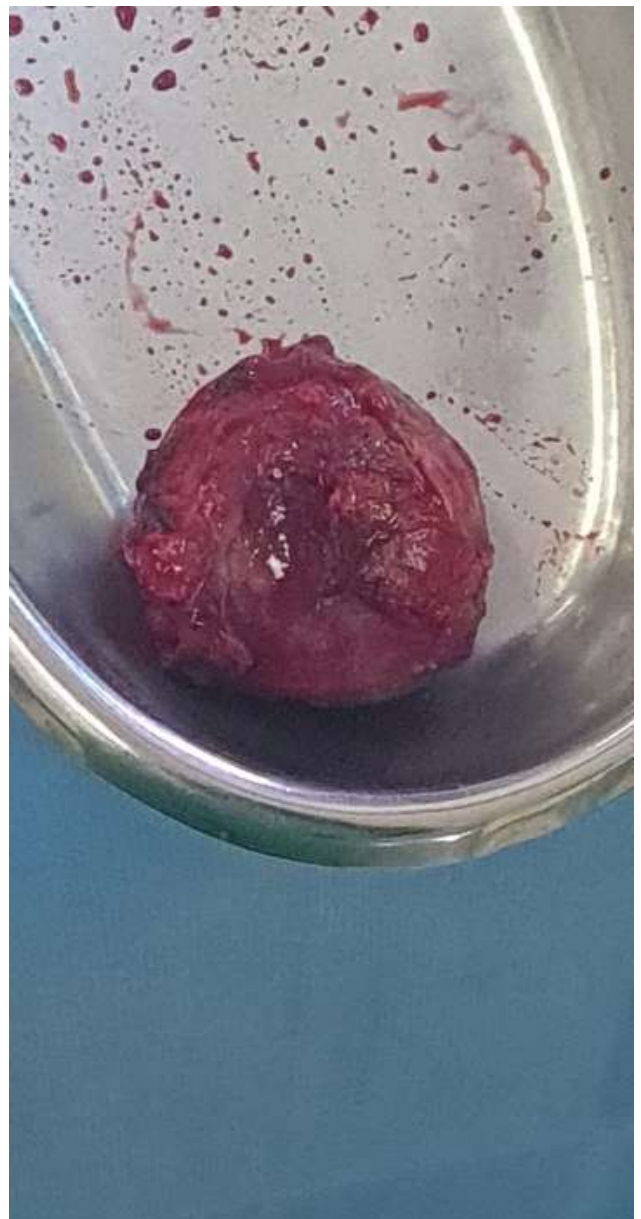


Figure 4: The mass after excision



The Surgical approach was a left subcostal incision.

A left encapsulated paraganglioma medial to the left ureter with adhesive bands to the aorta and anterior to the psoas muscles was found.

During the procedure when the mass was located and intraoperatively as the mass was gently manipulated for enucleation there were episodes of elevation of the blood pressure up to 180/70 mmHg and pulse rate to 118. At these moments the procedure was halted, with resumption only when the patient was normotensive.

Excision of the mass was successfully achieved with homeostasis. Examination of the adrenal gland and the kidney were done, abdominal wall closed in layers and skin incision closed with Monocryl subcuticular stitches.

## RESULT

Post operatively the patient was admitted in the high dependency unit for monitoring of blood pressure, pulse and fetal status. Her BPs were not erratic and remained normal throughout the post operative period. Below are the tabulated readings post procedure.

**Table 2: Recorded postoperative vital signs**

Blood pressure	Blood pressure	Pulse	FBS
Day 1 post op	158/94	112	4.3
Day 2 post op	143/84	100	8.2
Day 3 post op	108/56	113	5.2
Day 4 post op	132/77	86	8.9
Day 5 post op	127/67	102	7.2
Day 6 post op	139/88	97	7.2
Day 10 post op	118/67	84	5.0

Subsequently the vitals were noted to normalize apart from the pulse that kept on altering depending on the pain scale. The wound was exposed on day 3 post operatively and noted to have been well apposed without signs of surgical site infection. With resolution of most of the symptoms she was later discharged after 6 days to the ward with a viable fetus as evidenced by fetal movements and fetal heart rate. A post admission obstetric ultrasound was done that showed viable fetus.

Patient remained on oral Methyldopa, Nifedipine and labetalol with her BPS remaining normal. Her prior symptoms of headache, anxiety, diaphoresis subsided significantly.

Histopathology on the excised mass confirmed a para-aortic paraganglioma

At 33 weeks' gestation, an obstetric review revealed type 2 early onset fetal growth retardation due to placental insufficiency with brain sparing effect. Patient was immediately admitted, put on Magnesium sulfate, Dexamethasone and 8 hourly fetal monitoring. Based on the

fetus condition decision was made to deliver the fetus via caesarean section. Patient delivered a live male infant at 34 weeks, twelve weeks after her surgery, with Birth weight of 1206 grams, and immediately taken to the NBU unit under a neonatologist. Mother and child are currently discharged, child weighs 1315 grams on 34 mls of expressed milk and is doing well.

Her latest laboratory tests (20/4/2022) show that her 24HR Normetanephrine level at 2412 nmol/day (562-2129) down from a prep high of 36450, and a Normetanephrine: Creatinine ratio of 173.36umol/mol (50-204) also down from a prep level of 2934.

## DISCUSSION

Paraganglioma found in pregnancy are a rare occurrence by about 0.005%- 0.007% (1). Key importance is to manage the effects, as mortality of both the mother and the fetus have been noted to be high (45% and 77% respectively). With it relatively low incidence rates, the management is tailored to ensure that both the mother and the fetus are alive during and after pregnancy.

The management of BP in a pregnant woman with paraganglioma is a careful balance between providing adequate catecholamine blockade to reduce the risk of catecholamine excess while not compromising placental blood flow by lowering the BP excessively. In nonpregnant patients, a preoperative target BP of less than 130/80 mm Hg sitting but greater than 80/45 mm Hg standing has been recommended (9). It has been suggested that excessive BP lowering in pregnancy may result in uteroplacental insufficiency and intrauterine growth restriction, with a 176-g decrease in fetal weight with a 10-mm Hg decrease in mean arterial pressure in pregnant mothers without paragangliomas (10). In the context of pregnant women without paragangliomas, the threshold for treatment of hypertension in pregnancy varies, depending on which guideline is used, but treatment is suggested only if the BP is greater than 140–159 mm Hg systolic or 90–109 mm Hg diastolic (11). However, the issue with paraganglioma patients is the potentially labile BP with the risk of severe peaks of BP if no  $\alpha$ -blockade is given. Women with paragangliomas are at risk of placental abruption and fetal loss (7) which was almost the case with our patient. The fetus developed type 2 early onset fetal growth retardation secondary to placental insufficiency with brain sparing effect.

The choice of specific  $\alpha$ -blockade needs to be carefully considered in the management of paraganglioma in pregnancy. Phenoxybenzamine is an irreversible  $\alpha$ -adrenergic antagonist and has previously been suggested as the treatment of choice in women with paraganglioma or pheochromocytoma (12). Phenoxybenzamine has a half-life of 24 hours, and the clinical effects may continue for up to a week after discontinuation (13). Phenoxybenzamine has been shown to cross the placenta and accumulate in the fetus (8), potentially leading to neonatal respiratory depression and hypotension. As such, it has been recommended that neonates of women who have taken phenoxybenzamine in pregnancy are monitored for hypotension and respiratory depression in the first few days of life (14). It is theorized that the respiratory depression observed in these babies may result from an element of cardiac failure due to the large proportion of  $\alpha$ -adrenergic receptors in the fetal heart (14). Despite no teratogenic effects of phenoxybenzamine having been reported in animal testing, there are no well-controlled studies in pregnant woman. In the United States and the United Kingdom, phenoxybenzamine is a pregnancy category C, indicating that the pharmacological effects of the drug may cause harm to the fetus or neonate without causing

malformation. In our case, the baby was born with no birth defects, no neonatal respiratory distress or hypotension. We believe that close monitoring of the fetus and instituting dexamethasone, when the decision to deliver was made, aided in fetal lung maturation, and decreased the potential respiratory distress syndrome in this preterm delivered neonate (15).

Preoperative management with alpha blockers- phenoxybenzamine for 7-10 days before the procedure and further introduction a beta blocker, 3 days to the excision of the mass has been found to lower the blood pressure and reduced instances of intraoperative hypertensive crises (16). If the expectant mother has other comorbidities as in the case above, they should be well managed and controlled before the procedure. A thorough work up pre procedure should be done with an obstetrician, anesthesiologist, cardiologist, physicians, and the surgeon. A neonatologist should also be in the team, to potentially mitigate or deal with the possible complications of neonatal respiratory distress or hypotension

Traditionally it has been recommended that vaginal delivery is best avoided in pregnant women with pheochromocytomas or paragangliomas (17). There is a potential risk of precipitating a hypertensive crisis from active labor. There have been cases reported in the literature of successful normal vaginal deliveries without maternal or fetal mortality (18–24). However, in a case reported by Lyman (21), the postpartum period was complicated by pulmonary edema in the mother

Intraoperative management starts before induction of anesthesia where the patient is loaded with fluids as immediately post excision of the mass there is sudden decrease in the catecholamine secretions and the patients vital signs specifically the blood pressure may drop leading to other complications as acute ischemic attack, myocardial infarction and even death. Once the patient has been fluid loaded, she should be placed in the right lateral position where adequate Venous return can be achieved. Serial monitoring of the vital signs should also be done to maintain adequate vital signs. (25)

After the diagnosis of paraganglioma or pheochromocytoma in pregnancy, a multidisciplinary approach is essential. It is important that the mother with paraganglioma deliver in a tertiary hospital with an experienced obstetric, anesthetic, and endocrine service as well as a neonatal intensive unit. The delivery can be performed under a spinal anesthetic.

Spinal anesthetic may have a theoretical advantage of reducing neural stimulation to the adrenal glands and sympathetic chain if the block is high enough. It is also important to remember to avoid medications in these women that may precipitate a crisis, in particular agents such as metoclopramide (13). Ondansetron was successfully used in our case series without hemodynamic instability and has previously been reported to be used without adverse effects in metastatic pheochromocytoma (24).

A limitation with our case is the lack of genetic testing done for our patient. The study of familial paraganglioma/pheochromocytoma syndromes (also called hereditary paraganglioma/pheochromocytoma syndromes) has been very important in understanding the pathogenic mechanisms involved in both the familial as well as sporadic forms (25). Several susceptibility genes have been established as playing a central role in the pathogenesis of both pheochromocytomas and paragangliomas (26). Some of these represent inherited conditions (eg, pathogenic variants in the von Hippel-Lindau [VHL] tumor suppressor gene; the rearranged during transfection [RET] proto-oncogene; the neurofibromatosis type 1 [NF1] tumor suppressor gene; genes encoding for the four subunits [A, B, C, and D] of the succinate dehydrogenase [SDH] complex; a gene encoding the enzyme responsible for flavination of the SDHA subunit [SDHAF2]). Other pathogenic variants involve the gene-encoding transmembrane protein 127 (TMEM127), MYC-associated factor X (MAX), and hypoxia-inducible factor 2 alpha [HIF2A] (25). The highest malignancy rates are seen in paragangliomas associated with inherited pathogenic variants in the B subunit of the succinate dehydrogenase (SDHB) gene, which are usually abdominal and secretory.

Genetic testing, coupled with tumor location and tumor type aid in knowing the malignant potential and treatment protocol for patients with pheochromocytomas or paragangliomas. Further noting the rarity of paragangliomas in pregnancy, genetic testing, in this case, would have aided in adding to the already limited knowledge of managing such patients.

## CONCLUSION

Paragangliomas in pregnancy is very rare with maternal and fetal mortality rates higher than that of the general obstetric population, and a multidisciplinary approach with careful monitoring is highly recommended.

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SNK Waweru, S Opiyo, M Kanjumba participated in the management of this case and authored this manuscript. The final version of the text has been authorized for publication after revision by all the authors

## Conflict of interest

No conflict of interest

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## Penile strangulation by a metallic nut in a 10-year-old boy: a case report

**Mustafa O<sup>1</sup>, Hafswa A<sup>1</sup>, Syovata F<sup>2</sup>, Jumbi T<sup>2</sup>, Osawa F<sup>1</sup>, Yienya A<sup>1</sup>, Mburugu P<sup>2</sup>.**

<sup>1</sup>Department of Surgery, University of Nairobi

<sup>2</sup>Division of surgery, Kenyatta National Hospital

**Corresponding Author:** Dr Mustafa Omar. Email: musteeo@gmail.com

### ABSTRACT

**Background:** Penile strangulation is uncommon in children and is caused by circumferential constriction of the penile shaft by constricting material. Urologic surgeons frequently face tough hurdles while faced with penile constriction devices. Failure to remove such devices could result in serious ischemia and tissue loss. Patients frequently present after developing ischemia and edema for several days.

**Method:** A case report of a 10-year-old boy with a penile constrictive device who stayed for 6 days prior to presentation at a teaching and referral hospital in Nairobi, Kenya.

**Case discussion:** On arrival at the hospital the child was in pain and tachycardic. Local examination revealed an edematous hyperemic penile shaft that was entrapped at the base by a thick hexagonal metallic nut with penile skin ulceration and a discharge of straw-colored fluid. A diagnosis of penile strangulation by metallic nut was reached and an emergency removal under general anaesthesia was undertaken successfully using an electric angle grinder, a technique that has been used in adults in the same institution without complications. This child had no psychiatric illness.

**Conclusion:** Constrictive devices may result in serious injuries to the penis if not removed early. An electric angle grinder is a safe way of managing penile strangulation by a metallic nut in children.

**Keywords:** Penis, strangulation

## INTRODUCTION

Penile strangulation by foreign objects is not a common finding in paediatric urology. However, should it occur, it is a urologic emergency that requires immediate release of the strangulation so as to prevent outcomes of compartment syndrome, ischemia, necrosis, urethral injury, or even amputation of the penis. Although it's uncommon in children, it typically affects adults and teenagers because of erotic, autoerotic, enhancement of sexual performance, or occasionally because of devices used to collect urine in patients with enuresis or urine incontinence (1-3). Additionally, many patients also suffer from psychological illnesses (1, 2).

Penile strangulation may result from metallic, glass, plastic, or latex objects of all shapes and sizes (4–7). Rubber bands, threads, and hair are often the cause in children (1,8). Metal objects are typically difficult to remove and frequently cause penile injury, as opposed to non-metallic objects which are simple to remove (5).

Early removal of these objects from a strangulated penis is critical as it will prevent severe injuries. In many cases, the surgical approach for metallic objects requires industrial devices that are usually not readily available in urology theatres but nevertheless require the knowledge and competence to employ them (5).

In this article, we report our approach to a case of penile strangulation by a metallic nut in a 10 years old child which was successfully removed by an electric angle grinder. The authors recommend the electric angle grinder as a safe and effective technique for removing metallic nuts from a strangulated penis.

## CASE PRESENTATION

A ten-year-old boy was referred to Kenyatta National Hospital with a six-day history of penile swelling, pain and difficulty passing urine after inserting a metallic nut up to the base of his penis while experimenting. He claimed to have left it one day overnight and to have woken up with tumescence with the penis entrapped. The child persisted in trying to remove the ring, which caused the edema and penile shaft abrasion to worsen, and developed difficulty in passing urine. Due to embarrassment, the child continued to hide from the parents despite the pain getting worse. One day before admission, the pain became unbearable, and he was then brought to the hospital by his mother after noticing that his penis was grossly swollen and had a ring at the base. The rest of the history was unremarkable

On arrival at the emergency unit, the patient was in noted to be in pain and with a tachycardia of 118 beats per minute. He had no fever and was saturating at 97% on room air.

Local exam revealed an edematous penile shaft that was hyperemic and had an entrapment at the base of the penis by a thick hexagonal metallic nut. The penile skin was excoriated, with ulceration a discharge of straw-colored serous fluid from the skin (figure 1). The shaft was very tender on palpation and the urethral orifice was normal with no blood at the meatus. An abdominal exam revealed a distended bladder with tenderness. The rest of the systemic exam was unremarkable.

Figure 1: penile strangulation by metallic nut



Routine laboratory tests which included a haemogram and renal function tests were normal. Attempts at insertion of the urethral catheter were unsuccessful as the catheter could not advance past the membranous urethra. The patient was thus prepared for emergency examination under anesthesia and a plan to remove the ring.

In the theater, the patient was placed under general anesthesia and attempts to slide out the nut was unsuccessful even with lubrication. A ring cutter could not be used since the nut was too thick. Therefore, a decision to use an electric angle grinder was made due in view of the tough nature of the metallic nut (Figure 2).

Figure 2: **Electric Angle Grinder**



The nut was cut at 3 and 9 o'clock positions with artery forceps underneath the grinder as a barrier to protect the penile shaft (Figure 3). Continuous irrigation of the grinder blade with normal saline was necessary during the grinding process to prevent the production of sparks of heat that could cause burn injuries to the penis. After cutting for five minutes, a five-minute break had to be instituted to allow the heat to dissipate and the ring to cool down to avoid thermal injury to the patient. The process took approximately 30 minutes before the nut was totally removed after being sliced in two places.

Figure 3: **Grinder in use**



Thereafter, the penis was resuscitated with saline-soaked gauze. A 14fr silicone catheter was inserted under aseptic technique into the urethra and 1L of clear urine was drained. Adequate debridement of nonviable skin was done on the dorsal side (Figure 4). The penis was then cleaned and dressed in paraffin gauze.

Figure 4: **After removal of nut and resuscitation of penis**



The patient was put on a course of IV antibiotics and multimodal analgesic treatment postoperative. Daily wound management was carried out in the ward and the patient was discharged on postoperative day five after the removal of the catheter. Subsequent wound management was taken over by the aesthetic and reconstructive surgery team

## DISCUSSION

The risk of compartment syndrome and the loss of normal penile function, including erection and micturition, make penile strangulation a urologic emergency. Strangulation of the penis by foreign objects such as nuts is common among adults and adolescents. Most of these cases are as a result of the individual trying to enhance sexual performance (9). It is also common among individuals with psychiatric illnesses. The need for Psychiatric evaluation for such patients is controversial (11). It's uncommon in children and usually involves threads, rubber bands, and hair strands (13) (14) (12). It is mostly a result of experimenting and occasionally peer pressure from age mates.

Lymphatic and vascular congestion, ischemia, compartment syndrome, necrosis, with subsequent erectile dysfunction, and urethral damage defines the normal progressions of this strangling. If left untreated, necrosis with subsequent amputation of the penis is inevitable. Therefore, early intervention is warranted.

Due to humiliation, the child in this situation chose not to tell his parents what was happening. As a result, there was gradual edema and bladder outlet obstruction. It is difficult to remove these strangulating objects because doing so almost usually puts the penile shaft in danger of getting injured. Ring cutters, industrial wire cutters, pliers, and dental drills are just a few of the equipment that have been described in the literature as being used to remove these constrictive objects (1,10). In our case, we used an electric angle grinder due to the thickness of the nut involved. The risks of using this tool include injury to the penile shaft with possible amputation and thermal burns from the sparks released from the grinder in contact with the metallic nut. Another concern is that the grinder's operation on the metallic nut could cause thermal injury to the penile shaft.

To mitigate these risks, an artery forceps was placed between the nut and penile shaft to provide a barrier. A technique of continuous irrigation of the blade of the grinder with saline from a 20cc syringe during its operation was used to cool it down and prevent the sparks that would otherwise be generated. For every five-minute attempt at cutting the nut, a five-minute break was instituted to allow the ring to cool down as well as to prevent dissipation of the heat to the penile shaft. With these precautions in place, the nut was successfully removed without any complications. There was loss of skin on the dorsal penile shaft as a result of the prolonged edema and attempts at removal by the child that led to infection and excoriation.

Despite having the strangulation for a long time, our patient made a complete recovery with intact penile function. He however required skin grafting of the dorsal penile skin.

## CONCLUSION

Although it happens rarely in children, penile strangulation poses a major risk to the ability of the penis to function. In our case report, we used an electric angle grinder to remove a strangulating metallic nut. It is a viable option as long as the necessary precautions are taken to prevent further harm to the patient.

## Acknowledgments

Mustafa O , Syovata F, and Jumbi T participated in the management of this case and conceived of the presented report. Yienya A, and Mburugu P proposed the use of an electric angle grinder. Osawa F, and Mustafa O wrote the manuscript. All authors discussed the report and helped shape the final manuscript.

## Conflict of interest

No conflict of interest.

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# SISI PHARMACEUTICALS

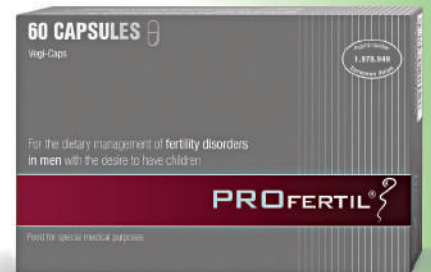
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