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Case Report

Robotic Excision of a Large Prostatic Utricle Cyst with Microdissection Testicular Sperm Extraction Presenting as Primary Infertility: A Case Report

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Abstract

Introduction: A prostatic utricle cyst is a rare congenital anomaly that appears as a midline diverticulum with its opening at the posterior urethra at the level of the verumontanum. The aim of this article is to present a rare case of a large prostatic utricle cyst as a cause of primary infertility, surgically managed with robotic excision followed by microdissection testicular sperm extraction to preserve fertility.

Case Presentation: We report a case of a 29-year-old male who presented with recurrent orchitis and primary infertility. Magnetic resonance imaging revealed an enlarged prostatic utricle cyst. Cystoscopy was performed to cannulate the cyst for guidance during laparoscopy. Robotic excision was successfully performed, followed by testicular extraction of motile sperm, which confirmed obstructive azoospermia secondary to the prostatic utricle cyst.

Discussion: Large symptomatic prostatic utricle cysts are managed surgically, whether open, endoscopic, laparoscopic, or robotic. Robotic-assisted laparoscopic excision of prostatic utricle cysts offers many advantages in terms of surgical field exposure, highly improved surgical dexterity and precision, fewer intraoperative complications, and postoperative outcomes. Testicular sperm extraction after surgery is recommended in such cases if fertility is desired.

Conclusion: Robotic excision with microdissection testicular sperm extraction for sperm cryopreservation for future fertility is a feasible and promising option for the treatment of large symptomatic prostatic utricle cysts.

Keywords: Prostatic utricle cyst; Robotics; Robotic surgery; Obstructive azoospermia; Primary infertility

Introduction

The prostatic utricle is a remnant of the Müllerian duct found in the posterior urethra of males. A Prostatic Utricle Cyst (PUC), on the other hand, is an abnormal dilation or enlargement of the prostatic utricle [1]. It is a rare entity in clinical practice, with a reported prevalence of 4% and 1% in children and adults, respectively [2]. When diagnosed, most cases are found concomitantly with other anomalies, such as cryptorchidism, unilateral renal agenesis, and hypospadias; more specifically, 11% - 14% are associated with distal hypospadias and disorders of sexual differentiation anomalies [3]. Diagnosis can be a clinical dilemma because most cases are asymptomatic and only appear during the first and second decades of life when signs and symptoms are present. This may include lower urinary tract symptoms, eventually leading to acute urinary retention, post-void dribbling, calculus formation, secondary incontinence, hematospermia, recurrent UTIs and epididymo-orchitis [4,5]. In rare instances, the presentation of primary infertility from obstructive azoospermia caused by a prostatic utricle cyst has been observed, with a reported incidence of approximately 1% in infertile males [2]. When there is a high index of suspicion, diagnosis is confirmed by various imaging techniques such as ultrasonography, retrograde urethrography, MRI, and direct visualization by cystoscopy, which is located at the posterior urethra near or at the verumontanum. It appears as a midline saccular structure situated between the urinary bladder and rectum. Symptomatic and large PUCs require surgical intervention wherein various techniques have been employed, including transurethral unroofing, open transvesical/perineal approaches, and laparoscopic excision [4]. Robot-assisted excision of PUC is becoming the technique of choice because it allows for better visualization of the relevant anatomy and manipulation of structures, leading to a safer and more satisfactory outcome [6]. We present the case of a 29-year-old male with recurrent epididymo-orchitis and obstructive azoospermia who was treated with robotic excision of a large prostatic utricle cyst followed by microdissection testicular sperm extraction. This case report has been reported in accordance with the SCARE criteria [7]. This study was granted a letter of exemption by the Institutional Ethics Review Committee Research and Biotechnology Group of St. Luke's Medical Center, Global City, Philippines (Reference No. SL - 24289).

Case Presentation

A 29-year-old Filipino male with a history of penoscrotal hypospadias, which was surgically corrected in two stages during infancy, had been experiencing recurrent orchitis and urinary tract infections for approximately 10 years. His symptoms included dysuria, incomplete voiding, increased urinary frequency, and recurrent scrotal fistulae. Initially, the patient responded well to

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antimicrobial treatment; however, his symptoms recurred frequently. The recurrent symptoms were initially attributed to a penile urethral stricture after hypospadias repair. The stricture was corrected by using a buccal mucosal graft. Despite correction of the stricture, the patient continued to experience recurrent tract infections and orchitis. A lower abdomen MRI was done in 2018 showing a large midline cystic structure occupying the entire prostate gland which measured 1.6 × 1.5 × 4.4 cm with no discrete focus of restricted diffusion suggesting a prostatic utricle cyst (Figure 1). The patient was advised to undergo surgical intervention; however, the family opted for maximum conservative management. One year later, the patient and his 26-year-old female partner were unable to conceive for a year. Semen analysis revealed the presence of azoospermia. The FSH, LH, and testosterone levels were within normal values at 7.72 mIU/ml, 6.25 mIU/ml, and 8.06 ng/ml respectively. Infection screening for sexually transmitted infections was negative. The patient consented to undergo robot-assisted laparoscopic surgical excision of the prostatic utricle cyst.

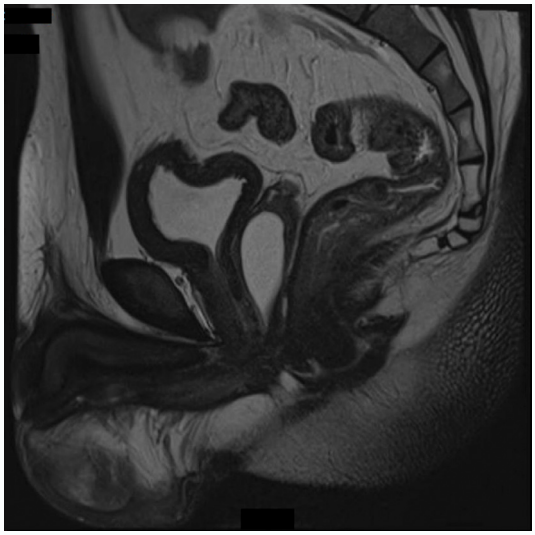

Figure 1: MRI T2-weighted sagittal view of the large prostatic utricle cyst showing its relationship to the rectum and urinary bladder.

On initial cystoscopy, the opening of the prostatic utricle cyst was located at the midline of the verumontanum. The verumontanum, which led to the prostatic utricle cyst, was cannulated using a French 5 open-ended catheter. A pediatric cystoscope was inserted to directly visualize and assess the cyst and its contents. An open-ended catheter was placed within the prostatic utricle cyst to facilitate intraabdominal identification (Figure 2). Other studies on the excision of prostatic utricle cysts described leaving a cystoscope within the cyst because the light at the end of the cystoscope served as a visual guide to its location as well as a way to manipulate the cyst intraabdominally during excision.

The 12 mm robotic camera port was inserted in the supraumbilical area, while two other ports were placed in each hemiabdomen to form a W configuration. A transabdominal posterior approach to peritoneal dissection was used to expose seminal vesicles and vas deferens. The bilateral vas deferens were carefully identified and were laterally inserted into the apex of the prostatic utricle cyst. This was also noted in two of our previous cases done laparoscopically. Additionally, because of its insertion, the vas deferens must be ligated to facilitate excision. The prostatic utricle cyst was circumferentially dissected and freed. Caudal dissection of the base of the prostatic utricle cyst eventually led to its insertion into the urethra. The prostatic utricle cyst was unroofed, revealing the previously inserted open ended catheter during cystoscopy, and was subsequently excised (Figure 3). Care was taken to leave at least a centimeter of the cyst to prevent injury and distortion of the urethra and bladder neck. The defect was repaired with continuous vicryl sutures. The excised specimen was delivered using a camera port. An indwelling Foley catheter was left in place after the procedure. Subsequently, laparoscopic ports were closed.

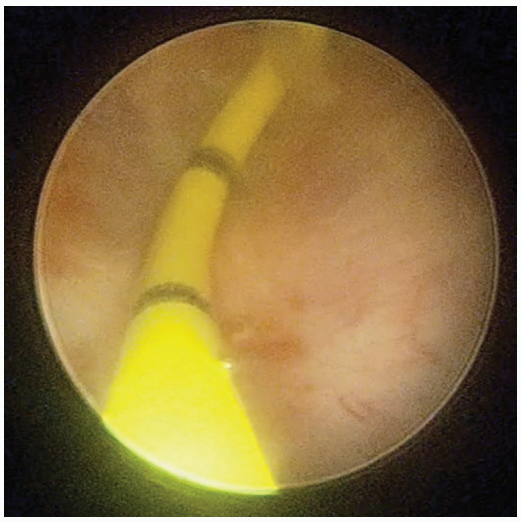

Figure 2: Cystoscopic view of the interior of the prostatic utricle cyst after cannulation of the opening using a 5 French open-ended catheter.

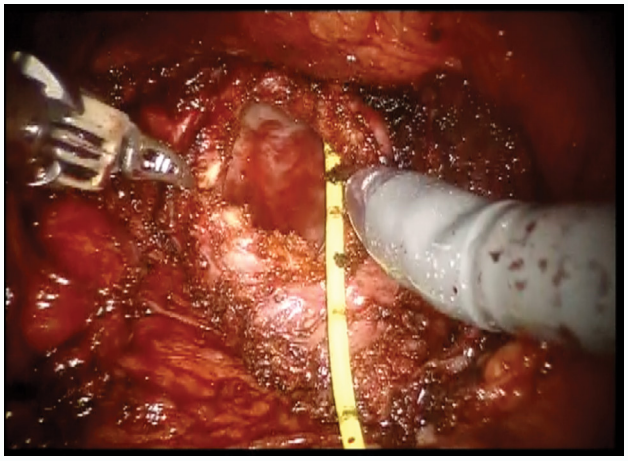

Figure 3: Laparoscopic view of the unroofed prostatic utricle cyst, showing the open-ended ureteral catheter inserted during cystoscopy.

Next, microdissection testicular sperm extraction was performed under microscopy. Healthy, motile sperm adequate for five to six *In Vitro* Fertilization (IVF) cycles were harvested. Biopsy revealed normal spermatogenesis and maturation in all seminiferous tubules, suggesting obstructive azoospermia from a prostatic utricle cyst.

The postoperative clinical course was uneventful. The patient was asymptomatic, had good pain control, and stable vital signs throughout the postoperative phase. Biopsy findings were consistent with those of a benign prostatic utricle. The patient was discharged

on 2nd postoperative day. The Foley catheter was maintained during discharge and subsequently removed after one week of follow-up. Incubation of sperm and subsequent fertilization of eggs *in vitro* produced five embryos ready for implantation into the uterus.

Discussion

Prostatic utricle cysts are vestigial saccular structures located at the posterior midline of the proximal half of the prostatic urethra which is a remnant of the Müllerian duct resulting from incomplete fusion during embryological development [8]. PUCs are different from Müllerian duct cysts in that the former have direct communication with the urethra, do not extend up to the prostatic base and are usually smaller in size compared to the latter measuring 0.8 cm - 1.0 cm on average [8]. PUCs result from the persistence of the fused caudal ends of the Müllerian duct during fetal development caused by reduced hormonal secretion and/or impaired hormonal sensitivity [1]. The functional testes of the developing embryo at 8 weeks produce Müllerian inhibiting factor facilitating regression of the Müllerian structures whereby a deficiency or dysfunction of the Müllerian inhibiting factor leads to the formation of PUCs. Histologically, PUCs arise from both mesodermal and endodermal tissues, forming a diverticulum wherein the cranial part is derived solely from the Müllerian duct, whereas the caudal portion comes from diversified tissues originating from both the Müllerian and Wolffian ducts, including the urogenital sinus.

PUCs are rare in the general population, occurring in 4% and 1% of infants and adults, respectively [2,9]. However, in the hypospadiac population, as in our patient, this was not an uncommon finding. It has a reported incidence of 14%, more specifically in the distal types [3,9]. Enlargement of the prostatic utricle cyst is directly correlated with the severity of penoscrotal hypospadias.

Most prostatic utricle cysts are asymptomatic and do not require surgical intervention. Typically, when symptoms are present, patients manifest during the first two decades of life [10]. If urine, semen, and debris are lodged in a hollow cyst, they can cause mass effects on the urethra, urinary bladder, and ureterovesical junction, leading to gradual pouch growth and cystic dilation. Bacterial contamination of the contents can lead to PUC symptoms. Documented complaints include irritative lower urinary tract symptoms such as post-void dribbling and urgency, urogenital tract infections presenting as recurrent epididymitis, and sexual dysfunction [1]. With an approximately 10-year history of recurrent epididymo-orchitis associated with primary infertility and voiding difficulty, our patient presented with all three groups of symptoms, wherein primary infertility was the main indication for surgical intervention. PUCs have been established as a risk factor for obstructive azoospermia leading to infertility, and two theories have been proposed. First, it is attributed to chronic genitourinary tract infections caused by PUC. Second, it is caused by compression or obstruction of the ejaculatory ducts by cystic dilation [4,8]. Preoperative sperm analysis revealed azoospermia, whereas intraoperative testicular sperm extraction revealed the presence of sperm cells, suggesting that the most likely cause of obstructive azoospermia may be the latter theory. In most cases, the vas deferens is attached to the prostatic utricle cyst and must be ligated, which results in male azoospermia. As a result, it is recommended that males who are undergoing excision of a prostatic utricle cyst where fertility is a concern, preoperative sperm banking, and cryopreservation must be offered. For cases in which the male is already azoospermic or severely oligospermic preoperatively, testicular sperm extraction plus cryopreservation may be offered to preserve the fertility potential.

Proper diagnosis of PUC is always a challenge, and most cases remain undiagnosed. PUCs are diagnosed incidentally on routine imaging such as ultrasonography, MRI, or cystourethroscopy [10]. Physical examination would typically be normal except when it is associated with intersex disorders and hypospadias, wherein a high index of suspicion will lead to further evaluation for the existence of a PUC. Until the mass is sufficiently enlarged, it may present as a palpable mass on digital rectal examination. Coupled with the clinical history, it may be easily confused with other differential diagnoses, such as ureteroceles, ejaculatory duct cysts, and prostatic retention cysts; however, it is difficult to distinguish it from Müllerian duct cysts. In contrast to Müllerian duct cysts, PUCs are most commonly pear-shaped and do not extend to the prostatic base. They have a direct communication with the prostatic urethra, with spermatozoa present and usually present earlier in life around the 1st or 2nd decade of life [8].

Urological imaging studies such as Retrograde Urethrography (RUG), Voiding Cystourethrography (VCUG), ultrasonography, Magnetic Resonance Imaging (MRI), and cystourethroscopy aid in the diagnosis of PUCs. The initial imaging modality is ultrasonography, which may reveal a midline dilated anechoic cystic structure posterior to the urethra and the urinary bladder. On MRI, because this structure contains fluid, it appears as high signal intensity on T2-weighted images, as depicted in Figure 1. Occasionally, when a PUC contains infected or hemorrhagic fluid, it may also be observed on MRI as a T1-weighted high-signal intensity [8]. Rarely used nowadays are invasive procedures, such as VCUG and RUG, which may also demonstrate a prostatic utricle cyst under fluoroscopic guidance. It is important to consider that through these modalities, the PUC may not be visualized, and diagnosis may be missed owing to incomplete filling of the cavity [11]. On cystoscopy, the opening can be visualized and cannulated at the posterior urethra, and a classification has been developed based on the opening of the PUC and the proximal extent of its cavity [11]. Based on this classification, our patient had a grade II prostatic utricle cyst because the opening was found in the posterior urethra, with its proximal body extending beyond the bladder neck and into the space posterior to the urinary bladder.

Large symptomatic PUCs require surgical intervention, and several techniques have been described, ranging from traditional open to more advanced robotic approaches. Surgical intervention is warranted to relieve the symptoms, preserve fertility, and rule out malignancy. Owing to its rarity in the clinical setting, anatomic degree of difficulty, and limited literature regarding the advantages and disadvantages of each approach, there is no consensus on the best approach [5]. Open surgical approaches, such as abdominal transperitoneal, perineal, anterior, and posterior sagittal transrectal techniques, have been described. These procedures require extensive pelvic dissection, poor exposure of the operative field, and incomplete PUC resection, thereby increasing the risk of vital pelvic organ damage [12]. A more recent technique, the suprapubic transvesical and extravesical approach, enables the surgeon to expose the bladder neck more effectively, allowing for safer dissection and complete resection of the PUC [5]. However, with the advent of laparoscopy and robotic surgery, open procedures have largely been replaced, and are currently not recommended. A study in 2016 compared the open transvesical technique with a laparoscopic approach and

concluded that the latter was more advantageous owing to its reduced postoperative pain, shorter hospital stay, and improved cosmetics [4]. Moreover, a modified cystoscopy-guided laparoscopic approach for countertraction of the PUC during dissection has been described and proven to ease surgeons throughout the procedure [12].

Here, we report a novel technique using robot-assisted laparoscopy to completely excise a prostatic utricle cyst. As in any other laparoscopic surgery, port placement was carefully planned to avoid instrument crossing. Robotic surgery allows even better exposure of the operative field owing to its refined Three-Dimensional (3D) visualization and a higher degree of magnification. Coupled with enhanced wristed instruments that provide high-end dexterity to surgeons, this technique leads to more promising intraoperative and postoperative outcomes [3]. Dissection of the cramped pelvic area is safer, with a lower risk of injury to surrounding structures. The improved robotic instruments allow for excellent precision and improved countertraction of the prostatic utricle cyst dissection. With the evolution of robotic surgery, assistive maneuvers in laparoscopy, such as transabdominal hitch stitches to elevate the pelvic structures and cystoscopy guidance for manipulation of the PUC intraoperatively, as previously mentioned, have been eliminated in robotic surgery, dramatically reducing operative time and inadvertent injury to associated structures [13]. In addition, with robotic surgery, minimal manipulation of the surrounding structures is required for maximal visualization, which is necessary in open procedures. At present, given the limited number of surgical cases of this rare clinical entity, the use of robotic surgery poses a steep challenge to urologists for its innovation, and further studies in the future might help determine the long-term outcomes of this technique.

Conclusion

Prostatic utricle cysts are rare congenital disorders in young males resulting from MIF deficiency, leading to incomplete fusion of the Müllerian duct remnants and failure of its obliteration. It is commonly associated with other congenital abnormalities, such as perineal or proximal hypospadias, unilateral renal agenesis, and disorders of sexual differentiation. The presentation varies widely; however, most patients are asymptomatic. Symptoms are brought about by gradual dilation of the cyst, and patients develop recurrent epididymoorchitis and obstructive azoospermia, posing a challenge to urologists. Large symptomatic PUCs can be managed surgically, whether open, endoscopic, laparoscopic, or robotic. Robot-assisted laparoscopic excision of PUC offers many advantages in terms of surgical field exposure, highly improved surgical dexterity and precision, and fewer intraoperative complications and postoperative outcomes.

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