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A Rare Case of an Inguinal Hernia-Containing (Extraperitoneal) Ureter

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Patient: Male, 67-year-old
Final Diagnosis: Extraperitoneal ureteroinguinal hernia
Symptoms: Fever • inguinal hernia • urinary frequency
Medication: —
Clinical Procedure: Hernioplasty • ureteral stent implantation
Specialty: Surgery • Urology

Objective: Rare disease
Background: Ureteroinguinal hernias are exceptionally rare and are seldom diagnosed in the preoperative setting. There are 2 classifications of this type of hernia: paraperitoneal and extraperitoneal.
Case Report: We report a case of a 67-year-old man who presented with urinary symptoms and a reducible right inguinal hernia. A computed tomography (CT) scan of the abdomen and pelvis suggested an ureteroinguinal hernia. Further diagnostics and treatment via cystoscopy, retrograde pyelogram, and right ureteral stent placement were performed, confirming the diagnosis and providing relief of the obstructive uropathy. The patient underwent an attempted elective transabdominal preperitoneal repair that was converted to an open Lichtenstein repair. Intraoperatively, an extraperitoneal ureteroinguinal hernia was identified. The patient did well postoperatively, and the stent was removed 1 month later.
Conclusions: Only 20% of the ureteroinguinal hernias described in the literature are extraperitoneal. In our case presentation, we demonstrated successful identification and treatment of an extraperitoneal ureteroinguinal hernia. The diagnosis was made using a combination of the clinical presentation, CT of the abdomen and pelvis, and cystoscopy with retrograde pyelogram. The extraperitoneal classification was an intraoperative diagnosis. The treatment consisted of a temporizing ureter stent and definitive management with an open Lichtenstein repair. We recommend obtaining a CT scan when a patient presents with a combination of urinary symptoms and an inguinal hernia because this process was invaluable in our preoperative diagnosis. Stent placement at the time of diagnosis permitted an elective repair and aided in the identification of the ureter during the hernia repair.

Keywords: Hernia, Inguinal • Herniorrhaphy • Ureteral Diseases

Full-text PDF: <https://www.amjcaserep.com/abstract/index/idArt/930911>



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Background

An inguinal hernia is a common ailment, with roughly 800 000 surgical repairs performed annually in the United States. However, finding a ureter located within an inguinal hernia is a rare discovery, with fewer than 150 cases reported in literature worldwide as of 2017 [1,2]. The available literature indicates that the majority of ureteral inguinal hernias are found at the time of operation and are less commonly identified in the preoperative period [3,4]. Awareness of the existence of such anatomic variation is important to prevent iatrogenic damage to the urologic structures during routine hernia repair [1]. Risk factors for ureteral inguinal hernias include male sex, age greater than 50 years, collagen synthesis deficiencies, history of a kidney transplant, and obesity [4,5]. Symptomatology may include dysuria, frequency, urgency, obstructive uropathy, 2-stage urination, nocturia, hematuria, ipsilateral flank pain, recurrent pyelonephritis, and incomplete emptying of the bladder. However, patients may be asymptomatic upon presentation [5,6].

Both computed tomography (CT) scan and ultrasound can provide reliable imaging to establish or confirm the diagnosis of a ureteroinguinal hernia [5]. However, the radiologic criterion standard for identifying a ureteroinguinal hernia is retrograde pyelography. The pathognomonic sign seen on the pyelographic evaluation is the “curlicue” or “loop-the-loop” sign. The presence of this sign is due to a redundant loop of the ureter and its abrupt changes in direction as it passes through the hernia defect [4,7]. There are 2 types of ureteroinguinal hernias described in the literature: paraperitoneal and extraperitoneal [3].

The majority of ureteroinguinal hernias are paraperitoneal; these are acquired and account for approximately 80% of ureteroinguinal hernias. They are defined by a ureter lying posterolateral to a peritoneal hernia sac, and they are generally accompanied by the herniation of other abdominal viscera. Paraperitoneal ureteroinguinal hernias are thought to arise due to adherence of the ureter to the hernia sac, which is then drawn into the inguinal canal as it follows the herniated viscera. They are usually large in nature, reducible, and asymptomatic [3,5].

Approximately 20% of the cases of ureteroinguinal hernias are the extraperitoneal type. They are congenital and identified by the lack of a peritoneal sac. This type usually herniates alone or in combination with retroperitoneal fat. Extraperitoneal ureteroinguinal hernias are due to the abnormal differentiation of the ureter from the Wolffian duct or adhesions to the genitinguinal ligaments that cause the ureters to descend into the scrotum with the testicles. These hernias are typically small defects, symptomatic, and nonreducible. They are frequently associated with renal or ureteral malformations such as renal ptosis, double-distinct ureter projection, and renal agenesis [3,5].

Case Report

A 67-year-old man presented to the Emergency Department with a 2-day history of suprapubic pain. The patient reported a history of suprapubic pressure, urinary incontinence, urinary urgency with frequency, and a fever of 39°C. He had a past medical history of tobacco use, congestive heart failure, diabetes, hypertension, postpolio syndrome, and obesity (body mass index=33.2 kg/m²). The patient had no prior abdominal surgeries. Physical examination identified a soft, nontender abdomen with reducible umbilical and bilateral inguinal hernias. Laboratory work revealed leukocytosis with a white blood cell count of 21 100/mm³. Blood urea nitrogen and creatinine levels were within normal limits. Urinalysis demonstrated a urinary tract infection with a moderate amount of leukocyte esterase and white blood cells. A CT scan of the abdomen and pelvis was performed per Emergency Department protocol, and it showed a right inguinal hernia containing a ureter, as well as hydronephrosis and ptosis of the right kidney consistent with obstructive uropathy secondary to an ureteroinguinal hernia. Imaging incidentally also revealed fat-containing left inguinal and umbilical hernias (Figures 1, 2).

The patient was subsequently admitted to the hospitalist service with consultation requested from Urology and General Surgery. He was started on antibiotic therapy, and cystoscopy with right retrograde pyelogram, and insertion of a right ureteral stent was performed to help alleviate his obstructive uropathy. Cystoscopy was performed using a 22F cystoscope, which was inserted into the bladder. The right ureteral orifice was identified and an open-ended catheter was used to perform the right retrograde pyelogram. The ureter was seen traversing the scrotum up to a dilated renal pelvis. The open-ended catheter was then passed through the intrascrotal ureter to the renal pelvis. The hernia was partly reduced to manipulate the catheter as it was traversing the ureter. Retrograde injection of contrast confirmed position in the renal pelvis. After the position of the renal pelvis was confirmed, a 035 stiff-shaft nitinol wire was placed in the renal pelvis. The open-ended catheter was removed. Using the Seldinger technique, a 6F by 32-cm variable-length stent was placed. Adequate coils were confirmed proximally in the renal pelvis and distally in the bladder. After ureteral stent placement, the patient had relief of symptoms, and the leukocytosis resolved. The patient was discharged from the hospital and elective herniorrhaphy was planned.

The patient returned to the operating room 2 months after the initial encounter. At this time, he underwent an attempted transabdominal preperitoneal (TAPP) hernia repair. The intraoperative evaluation identified a left-sided inguinal hernia. However, no peritoneal defect or ureter was identified on the right (Figure 3). Given this confounding finding, the operating



Figure 1. Coronal computed tomography scan of the abdomen and pelvis of a 67-year-old man with a ureteroinguinal hernia as well as right renal ptosis and hydronephrosis. The renal ptosis is apparent from the low-lying kidney seen in the image. The hydronephrosis is marked with an asterisk.



Figure 2. Sagittal computed tomography scan of the abdomen and pelvis in a 67-year-old man with ureteroinguinal hernia as well as right renal ptosis and hydronephrosis. The right ureter descending into a right inguinal hernia is visible in the image (arrow).

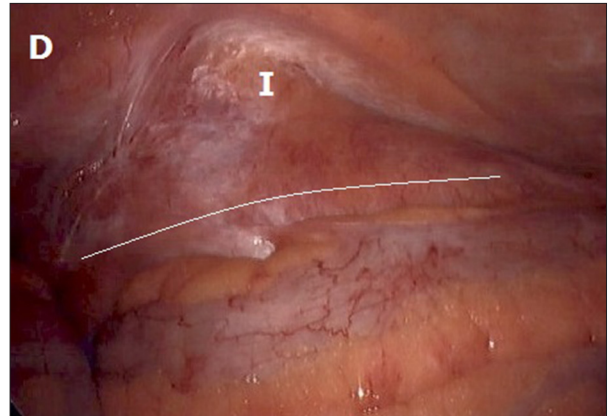


Figure 3. A laparoscopic image of the right peritoneum adjacent to the inguinal ligament (white line). Imaging demonstrated no peritoneal defect in the location where an indirect (I) or direct (D) hernia would be identified. No ureter was seen on laparoscopic evaluation. This lack of defect demonstrates evidence of an extraperitoneal ureteroinguinal hernia.

surgeon elected to perform an open repair. This decision was made due to inexperience with ureteroinguinal hernias and because an open approach is documented more frequently in the literature than other techniques for this type of hernia. Open dissection of the right inguinal region showed a large amount of retroperitoneal fat and the right ureter that had herniated from the retroperitoneum underneath an area of a direct inguinal hernia defect. The ureter was easily identified during the case secondary to the previously placed stent. The retroperitoneal fat and the ureter were reduced into the retroperitoneal space and the transversalis fascia was loosely re-approximated. A Lichtenstein mesh repair was then performed with a synthetic permanent mesh. Postoperatively, the patient underwent cystoscopy and right ureteral stent removal at follow-up 30 days later. The patient has had no postoperative complications to date.

Discussion

Ureteroinguinal hernias are extremely rare and as of 2017 fewer than 150 cases had been published in the literature [1]. Our report demonstrates both the diagnosis and the successful treatment of a rare extraperitoneal ureteroinguinal hernia. Our case involved a male patient with a reducible right inguinal hernia and urinary symptoms including suprapubic pressure, urinary incontinence, urgency, frequency, and obstructive uropathy. The diagnosis was established using a combination of clinical presentation, imaging, and operative findings.

Ureteroinguinal hernias are usually identified intraoperatively or postoperatively when an iatrogenic injury has occurred.

Identifying them in a preoperative setting is rare [1]. Based on clinical suspicion and advanced imaging, we were able to identify our patient's ureteroinguinal hernia in the preoperative setting. A literature review indicated that a CT scan can reliably establish the diagnosis of ureteroinguinal hernias [5]. According to recent literature, ureters displaced anteriorly from the psoas muscle by more than 1 cm at the level of L4 on CT scan may be predictive of ureteroinguinal herniation. In our patient, the ureter was located 1.27 cm from the body of L4 on CT scan, consistent with the measurement prediction by Allam et al [7]. Previous reports have demonstrated that extraperitoneal forms are frequently associated with renal or ureteral malformations such as renal ptosis, double-distal ureter projection, or renal agenesis [3-5,8,9]. A CT scan in our case suggested a possible extraperitoneal ureteroinguinal hernia due to the right kidney ptosis. However, the actual diagnosis of the specific form of ureteroinguinal hernia is an intraoperative finding [1]. Pareja-López et al [10] demonstrated that 3-dimensional CT scans yield a better understanding of the anatomy and aid in surgical decision-making [10]. Three-dimensional CT scan was not used in our case, but it may be beneficial in cases in which the anatomy remains unclear after obtaining a standard CT. The radiologic criterion standard for identifying a ureteroinguinal hernia is retrograde pyelography. As seen in our case, the pathognomonic sign on the pyelographic evaluation is the curlicue or loop-the-loop sign [4,7]. This sign can be seen clearly in **Figure 4**.

When a ureteroinguinal hernia is identified preoperatively, a multidisciplinary approach is recommended. Temporizing measures to prevent or treat obstructive uropathy should be performed, which often includes retrograde ureteric stent placement. At times the tortuosity of the ureter precludes standard stent placement and other means for ureteral drainage may be required, including angiocatheters or standard nephrostomy tubes [1,8,11]. In our case, a standard retrograde ureteric stent was placed despite the obvious tortuosity of the ureter. This allowed for the treatment of obstructive uropathy and also aided in the identification of the ureter during the definitive operation. Multiple reports have demonstrated that ureteric stents can also be beneficial in the localization of the ureter during the definitive herniorrhaphy [3,5,7,9].

The classic technique to repair ureteral hernias is an open preperitoneal, Lichtenstein, or Rutkow-Robbins hernioplasty [3,5,7,9]. Previous authors have encountered the impracticability of a pure laparoscopic approach and repair due to the anatomy [1]. Only as of November 2020 has the literature shown pure laparoscopic repair to be feasible [12]. Recently, laparoscopic robot-assisted techniques have been described and show promise [1,2]. The operating surgeon for our case attempted a pure TAPP laparoscopic repair despite prior literature questioning the probability of success. After encountering



Figure 4. The retrograde pyelogram shows the typical “curlicue” or “loop-the-loop” sign associated with a ureteroinguinal hernia in a 67-year-old man who presented with urinary symptoms and right inguinal hernia. A ureteral catheter is located in the distal ureter marked by an asterisk. Contrast leaving the catheter initially ascended into the abdomen before making a sharp turn and descending into the patient’s scrotum, where it made a loop and then ascended into the abdomen.

a lack of peritoneal defect, the operation was transitioned to an open repair. A totally extraperitoneal (TEP) repair is considered the standard technique of choice for most laparoscopic inguinal hernia repairs. However, this technique was not attempted in our case because the TAPP had a 2-fold advantage over the TEP repair in our case. The TEP repair can be challenging in patients who have a large abdominal wall pannus. Our patient was obese and had a pannus, thus a TEP repair would not have been ideal. Additionally, we have found through previous experience that there is improved ease of reduction of large hernias with a TAPP repair due to the larger working space. A TAPP repair allows access to the preperitoneal space after the peritoneum is incised. Had the operating surgeon opened the peritoneum overlying the inguinal hernia spaces, a defect would have been visualized and the CO₂ insufflated could have contributed to a spontaneous reduction of the ureteral hernia. No attempt at robotic repair was made owing to the unavailability of a robot platform at our institution. Regardless of the technique used, key steps for the procedure include careful dissection, ureteral replacement into the retroperitoneal space, and standard hernia repair. If the ureter is long and redundant, damaged, significantly dilated/tapered, or aperistaltic, or if it shows inflammation or necrosis, then it may require resection and reimplantation. Depending on the length of ureter involvement, options for repair include

ureteroneocystostomy, psoas hitch, Boari flap, or ureteroureterostomy [5,8,12]. There has been some literature that documents the utility of intraperitoneal fixation of the ureter to avoid ureteral volvulus [12].

Based on our experience, the preoperative CT scan was invaluable to identify the ureter in the inguinal canal and rule out any additional organ herniation. We would recommend obtaining a CT scan of the abdomen and pelvis in patients who present with urinary symptoms as well as an inguinal hernia. Our intraoperative identification of the extraperitoneal form and CT finding of renal ptosis support the correlation between renal defects and the extraperitoneal variant. Retrograde pyelogram with a ureteric stent aided in both changing the case from urgent to an elective status and allowed for the identification of the ureter intraoperatively. Therefore, we continue to recommend its use if the hernia is identified preoperatively. In our case, we attempted a transabdominal laparoscopic inguinal hernia repair. The lack of identification of a peritoneal hernia defect on the right and a review of prior literature led to our decision to convert to an open approach. What we have learned from further review of the literature is that a peritoneal defect may not be present based upon the pathophysiology of the extraperitoneal ureteroinguinal hernia. If the hernia had been in the paraperitoneal form, a peritoneal defect would have been visualized. In our case, if we would have excised the peritoneum and gained access to the preperitoneal space, a defect would have been visualized, and had we seen the defect, we may have considered continued attempts at laparoscopic repair at that time. A recent case report describes the first successful laparoscopic repair of an extraperitoneal ureteral inguinal hernia and can be used as a resource for future planned repairs of ureteral inguinal hernias [12]. A review of ureteroinguinal hernias and surgical approaches to standardize the treatment would be an interesting focus of further research on this topic.

References:

1. Pucheril D, Chun B, Dalela D, et al. Robot-assisted laparoscopic repair of extraperitoneal ureteral inguinal hernia with mesh placement. *J Endourol Case Rep.* 2017;3(1):97-100
2. Cetrulo LN, Harmon J, Ortiz J, et al. Case report of a robotic-assisted laparoscopic repair of a giant incarcerated recurrent inguinal hernia containing bladder and ureters. *Int J Med Robot.* 2015;11(1):15-17
3. Bertolaccini L, Giacomelli G, Bozzo RE, et al. Inguino-scrotal hernia of a double district ureter: Case report and literature review. *Hernia.* 2005;9(3):291-93
4. Malde S, Bilagi P, Marsh H. Obstructive uropathy due to uretero-inguinal hernia: An uncommon occurrence. *Indian J Urol.* 2013;29(4):355-56
5. Morrison Z, Nirujogi VL. Ureteral inguinal hernia. *Treasure Island (FL): StatPearls,* 2008
6. Ahmed S, Stanford R. Ureteric obstruction secondary to a paraperitoneal inguinal hernia. *Ann R Coll Surg Engl.* 2016;98(2):e16-18
7. Allam ES, Johnson DY, Grewal SG, Johnson FE. A sign on CT that predicts a hazardous ureteral anomaly. *Int J Surg Case Rep.* 2016;22:51-54
8. McKay JP, Organ M, Bagnell S, et al. Inguinoscrotal hernias involving urologic organs: A case series. *Can Urol Assoc J.* 2014;8(5-6):E429-32
9. Osmani HT, Boulton R, Wyatt H, Saunders SM. An unusual inguinoscrotal hernia with renal involvement. *BMJ Case Rep.* 2015;2015:bcr2015211519
10. Pareja-López Á, Sevilla-Cecilia C, Pey-Camps A, et al. Ureteroinguinal hernia. *Cir Esp.* 2015;93(8):e89-90
11. Feyisetan O, Floyd MS Jr, Samsudin A. Obstructive uropathy secondary to bilateral ureteroinguinoscrotal herniation. *Int Braz J Urol.* 2016;42(3):621-22
12. Narayanan PL, Narayanan CD, Sekar V, Vadyala AR. Laparoscopic approach to ureteroinguinal hernia. *Int J Surg Case Rep.* 2020;77:161-64

Conclusions

A ureteroinguinal hernia is an extremely rare finding. The literature describes 2 types of ureteroinguinal hernias, paraperitoneal and extraperitoneal, with the latter only accounting for approximately 20% of the cases. Ureteroinguinal hernias may present with characteristic urinary symptomatology and obstructive uropathy, or they may be asymptomatic. Based on clinical presentation, CT findings, retrograde pyelogram, and operative findings, we diagnosed a rare case of an extraperitoneal ureteroinguinal hernia. The obstructive uropathy was successfully treated with an urgent ureteral stent, and the hernia was treated with an elective Lichtenstein mesh repair after an aborted laparoscopic TAPP repair. Based upon our experience with this rare form of hernia, we can make some recommendations. A preoperative CT scan was invaluable to identify the ureter in the inguinal canal and rule out any additional organ herniation. Retrograde pyelogram with ureteric stent aided in both changing the case from urgent to elective status and assisted in the intraoperative identification of the ureter. While we attempted to perform TAPP, the lack of identification of peritoneal defect on the right side confounded our understanding and planned approach. What we have learned in reviewing the literature is that, based upon the pathophysiology of an extraperitoneal ureteroinguinal hernia, a defect will not be seen from an intraperitoneal approach; however, it would be identified if it is a paraperitoneal type. Additionally, if the peritoneum had been incised or we had performed a laparoscopic TEP, then the defect would have been visualized and potentially repaired using the laparoscopic technique.

Conflict of Interest

None declared.

Declaration of Figures Authenticity

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