A COMPLETE DUPLICATED SYSTEM WITH INTRAVESICAL URETEROCELE IN A 60-YEARS-OLD WOMAN: A CASE REPORT

[1] Satria Adji Hady Prabowo, [1]Taufiq Nur Budaya, [1]Andri Kustono.

[1] Department of Urology, Faculty of Medicine/University of Brawijaya, Saiful Anwar General Hospital, Malang.

ABSTRACT

Objective: *This case report presents a 60-year-old woman with intravesical ureterocele complicated by severe right upper-moiety hydronephrosis.****Case(s) Presentation:*** *A 60-year-old woman came with the chief complaint of right flank pain radiating to the umbilicus and groin accompanied by nausea, vomiting and decreased appetite. Ultrasound examination showed severe hydronephrosis of the right kidney. Abdominal CT revealed a double collecting system with severe hydroureteronephrosis at the upper pelvicalyceal system and the presence of an intravesical ureterocele. Cystoscopy and right RPG of upper and lower moiety procedures were performed and showed right severe hydroureteronephrosis with stenosis of right upper moiety ureteral orifice and ureterocele, followed by uroterocele incision and upper moiety ureteral re-implantation. No further complaints or complications were found after discharge from the hospital.* ***Discussion****:Adult ureterocele is a rare congenital anomaly often detected incidentally or due to complications like obstruction and infection.Diagnosis relies on imaging modalities such as ultrasonography, intravenous urography, and CT urography, while treatment options range from minimally invasive endoscopic decompression to surgical excision, depending on severity and associated complications.* ***Conclusion:*** *Ureterocele is a rare congenital urological abnormality found in non-Caucasians. Due to the rarity of the case, a thorough examination is required to establish this dia\gnosis and the choice of the appropriate treatment procedure. The managements include incision of ureterocele and ureteroneocystostomy.*

Keywords: *Adult ureterocele,intravesical ureterocele, hydronephrosis, complete double collecting system, vesicoureteral reflux*

ABSTRAK

Tujuan: *Laporan kasus ini membahas seorang wanita 60 tahun dengan ureterokel intravesika yang disertai hidronefrosis berat pada moiety superior kanan.* ***Presentasi Kasus:*** *Seorang wanita berusia 60 tahun datang dengan keluhan utama nyeri pinggang kanan menjalar ke pusar dan pangkal paha disertai mual, muntah dan nafsu makan menurun. Pemeriksaan ultrasonografi menunjukkan hidronefrosis berat pada ginjal kanan. CT abdomen menunjukkandouble collecting system dengan hidroureteronefrosis berat pada sistem pelviokalises atas dan adanya ureterocele intravesikal. Sistoskopi dan RPG kanan prosedur bagian atas dan bawah dilakukan dan menunjukkan hidroureteronefrosis berat kanan dengan stenosis muara ureter bagian atas kanan dan ureterocele, diikuti oleh insisi uroterocele dan re-implantasi ureter bagian atas. Tidak ditemukan keluhan atau komplikasi lebih lanjut setelah keluar dari rumah sakit.* ***Diskusi:*** *Ureterocele pada dewasa merupakan kelainan kongenital yang jarang terjadi dan sering terdeteksi secara insidental akibat komplikasi seperti obstruksi dan infeksi. Diagnosis bergantung pada modalitas pencitraan seperti ultrasonografi, urografi intravena, dan CT urografi, sementara pilihan pengobatan bervariasi dari dekompresi endoskopi minimal invasif hingga eksisi bedah tergantung pada tingkat keparahan dan komplikasi yang menyertainya.****Simpulan:*** *Ureterokel merupakan kelainankongenital dibidang urologi langka yang ditemukan pada orang non-Kaukasia.Karena kasus ini jarang terjadi, diperlukan pemeriksaan menyeluruh untuk menegakkan diagnosis ini dan memilih prosedur pengobatan yang tepat. Penatalaksanaannya meliputi insisi ureterokel dan ureteroneosistostomi.*

Kata kunci: *Ureterokel dewasa, ureterokel intravesikal, hidronefrosis,double collecting system, refluks vesicoureteral*

Correspondence: Taufiq Nur Budaya; c/o: Department of Urology, Faculty of Medicine/University of Brawijaya, Saiful Anwar GeneralHospital, Jl. Jaksa Agung Suprapto No.2, Klojen, Kec. Klojen, Malang, Jawa Timur 65112, Indonesia. Phone: +6281217177393. Fax: +62341333030. Email: Taufiq_fkub03@yahoo.com.

INTRODUCTION

Ureterocele is a congenital abnormality with cystic dilatation of the lower ureter. These abnormalities can cause various effects related to obstruction, reflux, continence, and renal function disorder.[1] Ureteroceles often present with other urologic abnormalities such as ureteral orifice

stenosis or a duplex collecting system.[2] Duplicated collecting system, defined as a kidney with two pelvicalyceal systems, which may have a single ureter or bifid (partial duplication) or multiple ureters draining separated independently into the bladder (complete duplication).[3] Ureteroceles can be classified as intravesical or extravesical ureterocele.[1] Ureteroceles are more common in children or women and usually manifest as infection and abdominal pain.[4] This condition may not be recognized until adulthood. Ureterocele is usually found on radiological examination.[5] This case is presented because it is rarely found in non-Caucasians, especially in this case it occurs in Asian individuals who come from Indonesia.

Ureterocele is one of the urological disorders that is challenging to be faced by pediatric and adult urologists because of the prevalence of cases occurring in 1:4000 children in America and Europe, even 4-7 times more common in women with the majority found in white or Caucasian patients with various types of clinical presentation.[5-6] When diagnosed in adulthood, the most common type of ureterocele is the intravesical type, rarely causes obstruction, and is usually associated with a single system.[7] In this case report, we will discuss a case of intravesical ureterocele complicated by severe right upper-moeity hydronephrosis in a patient. 60 year old woman.

CASE PRESENTATION

A 60-year-old woman came with the chief complaint of right flank pain for 10 months, worsen in the last 1 month. The pain radiates to the umbilicus and right groin. Pain exacerbates with activity, and does not improve with analgesics. Patient also complained of nausea, vomiting and decreased appetite since the last 2 weeks. Patient had been diagnosed with diabetes mellitus since 3 years ago, routinely consumed Glibenclamide 5 mg once a day since 1 year, the patient also had history of cholecystectomy. On physical examination, we found right costovertebral angle flank pain, with spontaneous micturition and normal laboratory findings.

Ultrasound examination showed severe hydronephrosis of the right kidney (Figure 1). On abdominal CT Scan examination, we found a double collecting system with severe dilation of the upper pelvicalyceal system to the ureterovesical junction with protrusion of the distal end of the ureter into the bladder, with the size of 4.6 x 4.8 x 3.3 cm. Dilated, tortuous ureters and thickening of the upper moeity ureteral wall were seen at L5-S1 level (Figure 2).

Cytoscopy and RPG performed with result (Figure 3; Figure 4), an ureterocele at the right side of bladder, inferior to normal ureteral orifice, we also found ureteral orifice stenosis at upper moeity.

The procedure followed by the incision of ureterocele and RPG. After ureterocele incision, we found severe hydroureteronephrosis in the upper moiety of right kidney (Figure 4). After that, we continue with right upper moiety ureteral re-implantation (Figure 5). The DJ stent was removed one month postoperatively.

At the six-month postoperative follow-up, the patient presented to the outpatient clinic with a well-healed surgical wound. A urological ultrasound was performed and revealing no hydronephrosis in the right kidney. The patient reported no significant complaints.

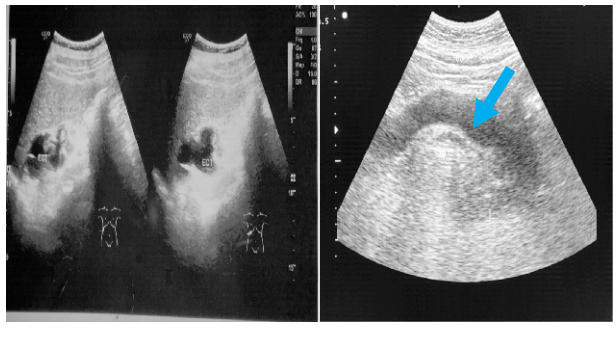

Figure 1. Abdominal Ultrasound shows (A) Severe Hydronephrosis of Right Kidney(B) Intravesical Ureterocele (blue arrow).

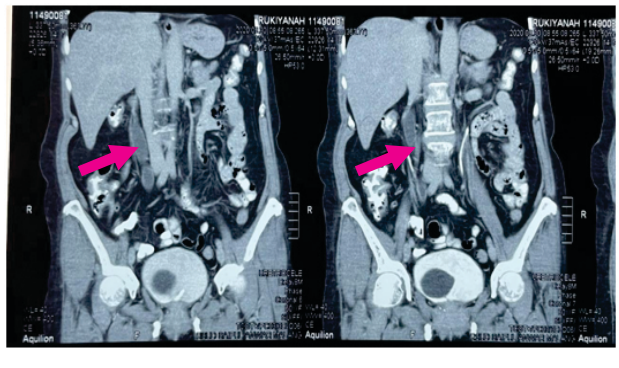

Figure 2. Abdominal CT Scan with Contrast. Duplex collecting system with right upper severe hydroureteronephrosis (red arrow) with intravesica ureterocele (purple arrow).

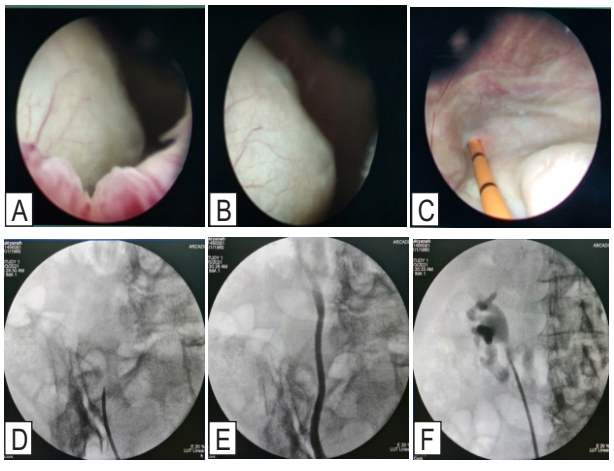

Figure 3. Cystoscopy and RPG Examination of Right Lower Moiety. (A-B) Uroterocele in the right bladder wall (C) novel ureteral orifice just proximally from ureterocele; (D-F) RPG examination: lower moiety is normal

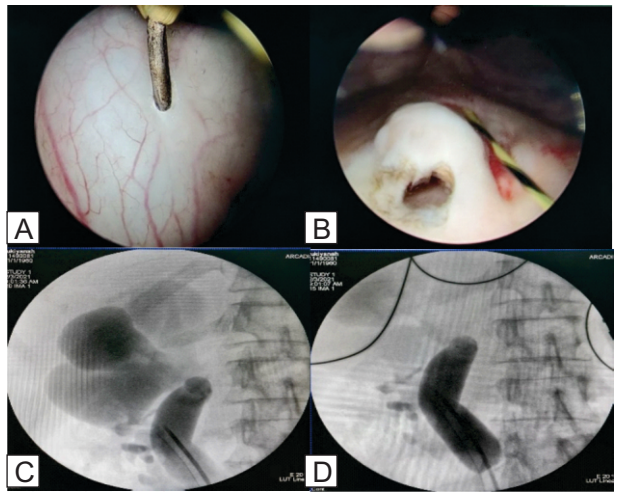

Figure 4. Ureterocele Incision and RPG Examination of right Upper Moeity. (A-B) Right ureterocele incision; (C-D) RPG to upper moiety: right severe hydroureteronephrosis.

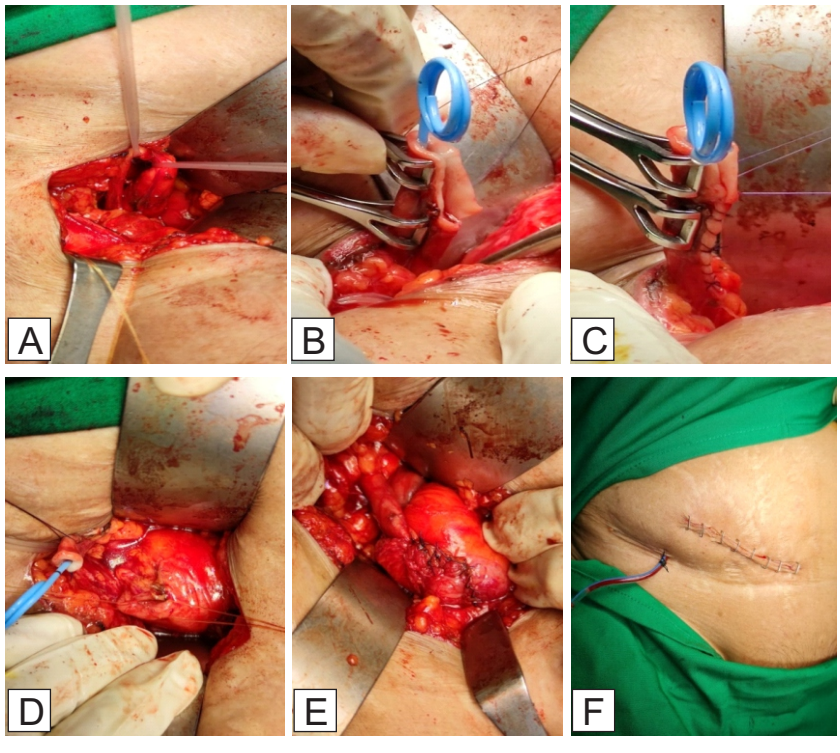

Figure 5. Ureteroneocystostomy Procedure. (A) Gibson incision to midline 8cm long. The peritoneum is dissected medially, identify the upper moiety ureter, and perform cranial and caudal release and hold the ureter, then evaluate and fix the bladder; (B-C) The ureter is cut as distally as possible and the distal part is sutured; (D) Incision of the bladder to the mucosa and placement of a DJ stent; (E) A ureteroneocystostomy technique was performed with six sutures. After that, the anti-reflux procedure was carried out, followed by (F) washing the operating layer, installing an 11 Fr drain, done with closing and suturing layer by layer.

DISCUSSION

Ureterocele is a cystic dilatation of the sub-mucosal segment of the intravesical ureter, and its etiology remains uncertain. It is commonly unilateral, but is bilateral in 10% of cases. [8-9] Ureteroceles are generally considered congenital. Failure of the Chwalla membrane to completely perforate during development of the ureteral orifice is thought to explain the occurrence of a ureterocele. However, inflammation or trauma can also cause the development of an orthotopic ureterocele from fibrosis around the ureteral orifice.[10]

A ureterocele is a well-known entity among pediatric urologic population but it remains a challenge in the adult population. Most of our knowledge is based on case reports and case series. Based on autopsy studies, its prevalence in the adult population ranges between 1/500 and 1/4000. We tried to search PubMed using the terms "adult ureterocele", "ureterocele in adult". In Indonesia, we find one ureterocele case report in adult reported by Yuri et al.[2]

There are multiple classification systems such as the Stephens classification which depends on the size and location of the ureteric orifice or the functional based classification by Churchill, however due to their complexity these systems have gained less popularity and more simplified system was established by the American Academy of Pediatrics (APA).[1] APA proposed standardized terms of ureterocele. They include: intravesical ureterocele (ureterocele contained wholly in the bladder), ectopic if any portion extends to the bladder neck or urethra, single or duplex systems and in term or the orifices as terms stenotic, sphincteric, sphincterostenotic, and cecoureterocele were recommended.[8] Single-system ureterocele is usually found in adults, and are almost always intravesical. It is less prone to obstruction and renal dysplasia.[8,11]

Adult ureteroceles are less prone to obstruction and dysplasia, and thus are generally asymptomatic. Symptoms of ureterocele can include lower urinary tract symptoms, flank or pelvic pain, and hematuria. This explains why they are detected in adulthood either incidentally or when a patient presents with recurrent flank pain, calculus, or UTI.[8,11] In this case, the patient presented with right flank pain radiating to umbilicus and right crotch.

Ultrasonography was the first imaging technique used in the evaluation of a ureterocele. Ultrasonography can confirm the diagnosis, because a cystic mass may be seen in the VU or near the proximal urethra. Intravenous urography may show poor function on the affected side with delayed or no excretion. Examination may also show hydroureteronephrosis, a dilated distal ureter, presenting as a "cobra head" or "spring onion" deformity with a peripheral halo. These findings are recorded in intravenousurography (IVU).[10] Preoperative imaging with contrast abdominal CT or CT urography (CTU) can also be useful in demonstrating structural abnormalities of the urinary tract, determining the size, shape, and location of the ureterocele.[12] Ureteroceles may be seen as cobra-head appearance. A double collecting system is usually indicated by a prolonged delay in excretion at the CTU due to a malfunction in dispensing contrast with the duplex ureter. Cystoscopy can be performed routinely or selectively.[6,11] Intraoperative retrograde pyelography or retrograde pyelography (RPG) can be used to define the anatomy of the ureterocele and to assess the presence of vesicoureteral reflux or vesicoureteral reflux (VUR)[13]. In this case, ultrasound and CT scan of the abdomen with contrast showed complications of ureterocele in the form of severe right hydronephrosis and intravesical ureterocele.

Inthis case,severehydroureteronephrosis and stenosis of right upper moiety ureteral orifice was found as the complication of ureterocele. Ureterocele in a duplex system most commonly involves the upper pole moiety and hydronephrosis in the upper moiety is usually due to the obstruction at the lower end while in the lower moiety it is usually due to the vesicoureteric reflux.[14] There are few case reports showed ureterocele complicated by hydronephrosis. Ileri et al reported incidental ureterocele complicated with hydronephrosis in adult.[11] Also Vasu et al and Dada et al reported a patient with a progressed disease to renal failure due to bilateral ureterocele.[5,15,16]

The managements generally aim to prevent urinary tract infections, maintain renal function, ensure good ureterovesical drainage, prevent vesicoureteral reflux, minimize surgical complications with a minimum number of procedures and to maintain the possibility of further reconstruction. Several different techniques (eg, incision and resection) have been performed to decompress ureterocele in adults. The endoscopic approach, with the benefits of its short operative time and good clinical outcome, is considered the gold

standard for intravesical ureterocele. The goal of the endoscopic technique is to decompress the obstructed system minimizing the incidence of postoperative reflux. Other procedures such as upper pole nephrectomy or excision of the ureterocele and common sheath reimplantation are required based on the renal function of the site involved, persistent obstruction or vesicoureteral reflux of the upper tract and the occurrence of new post-procedure vesicoureteral reflux.[9,10,17]

Ureterocele management depends on factors such as clinical presentation, age, ureterocele type, infection, stones, and duplex kidney involvement. Surgical options include incision, punctures, unroofing, and resection, with endoscopic procedures being a preferred minimally invasive approach for early decompression. Studies confirm their safety and efficacy.

Although previous research indicates that minimally invasive surgery often yields positive outcomes without symptom recurrence, certain cases require secondary intervention following an initial procedure.[18] Specifically, in patients with ectopic ureteroceles and duplex systems with pre-existing reflux, endoscopic incision alone may not provide definitive treatment.[19] Nevertheless, the overarching goals of ureterocele management is for preventing urinary tract infections, relieving obstruction of renal parenchyma, managing vesicoureteral reflux, and minimizing surgical morbidity.[20] In previous case, the patient underwent endoscopic ureterocele unroofing, a minimally invasive procedure to relieve obstruction and improve urinary flow. The patient recovered well without postoperative complications.[21]

A previous case report highlighted the role of open surgical repair in managing ureterocele, particularly in resource-limited settings. In that case, the patient consented to open surgery, revealing a large right ureterocele (6 cm × 3 cm) protruding into the bladder, while the left ureteric orifice and bladder wall appeared normal. The procedure involved marsupialization after partial excision of the redundant ureterocele wall. A temporary ureteric stent was removed two weeks postoperatively, and the patient showed good recovery at the six-month follow-up.[22]

The patients in this case underwent surgical treatment with ureterocele incision to alleviate hydroureteronephrosis, enhance kidney function, and lower the risk of urinary tract infection. Additionally, right upper moiety neoimplantation was performed to prevent vesicoureteral reflux (VUR) and further reduce the risk of urinary tract infections.

This study has limitations included this case report focuses on a single patient, which restricts the extent to which the findings can be generalized. Although it offers useful clinical perspectives on the diagnosis and treatment of adult ureterocele, the results may not fully represent broader patient populations. Moreover, the follow-up period of six months is relatively short, limiting the assessment of potential long-term complications or recurrence. Further research with larger sample sizes and extended follow-up is required to confirm the efficacy of the surgical approach and refine management strategies for adult ureterocele.

CONCLUSION

Adult ureterocele is a rare condition, primarily of the intravesical type, as observed in this case. It is often diagnosed incidentally or when patients present with recurrent low back pain. Due to its variable clinical presentation, diagnosis can be challenging and typically requires imaging studies such as ultrasound, CT scan, and retrograde pyelography. In this patient, surgical intervention included ureterocele incision and ureteroneocystostomy. Given the rarity of this condition, a thorough and individualized approach is necessary for effective management. Long-term follow-up remains crucial to detect symptom recurrence or potential complications

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