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Multiple stones in a pediatric case of single-system ureterocele with vesicoureteral reflux

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ABSTRACT

Presence of multiple calculi in a single system ureterocele is a rare condition. A 3-year-old boy presented with recurrent urinary tract infections in whom multiple calculi were noted in the urinary bladder on x-ray and ultrasound scan. In addition, ultrasound showed the presence of ureterocele in the size of 18x15x12 mm. Open surgery revealed an ureterocele with multiple stones. Here, we present a boy with multiple stones in the left ureterocele diagnosed intra-operatively due to its rarity, etiology and treatment options.

Key Words: Ureterocele, stones, children.

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Introduction

Ureterocele is defined as the cystic dilatation of the distal ureter that is thought to be associated with bladder-derived tissue defect [1]. It is a congenital anomaly of the distal ureter and is seen in approximately 1/4000 live births [2]. The pathology can be seen with a single or duplicated collecting system. The ureterocele may settle as orthotopic [intravesical] or ectopic relative to it's opening in the bladder. Delay in the diagnosis of ureterocele cases can lead to severe complications from recurrent urinary tract infections (UTI) to renal failure [2]. In addition, multiple calculi in a single system ureterocele is a rare entity in the literature. In this case report, we aimed to present a case of ureterocele with urinary stone in a 3-year-old boy and to present the etiology and treatment options of this uncommon condition.

Case report

A 3-year-old male patient was admitted with a complaint of recurrent UTI. Physical examination was normal except for suprapubic sensitivity. The urine culture was negative. Ultrasonography (US) showed a view of ureterocele with an 18x15x12 mm size and a large number of stones up to seven millimeters in it with acoustic shadow on the left posterolateral side of bladder (Fig. 1). Voiding cystourethrogram (VCUG) was performed due

to recurrent UTI. Left Grade 1 vesicoureteral reflux (VUR) was detected (Fig. 2). The kidney-ureter-bladder radiography showed several clustered opacities of approximately 4-5 mm in the bone pelvis on the left side (Fig. 3).

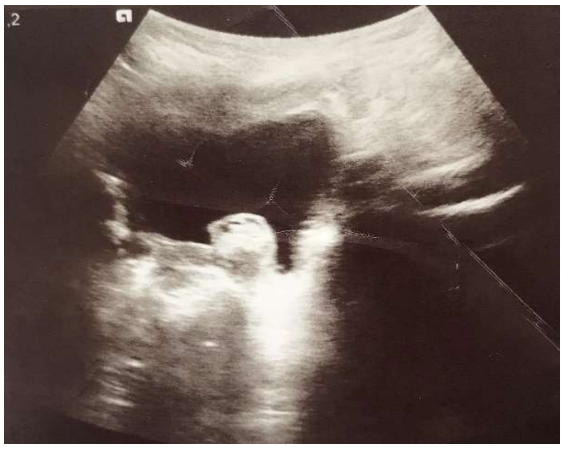

Fig. 1. Ultrasonography image of the stone in the ureterocele.

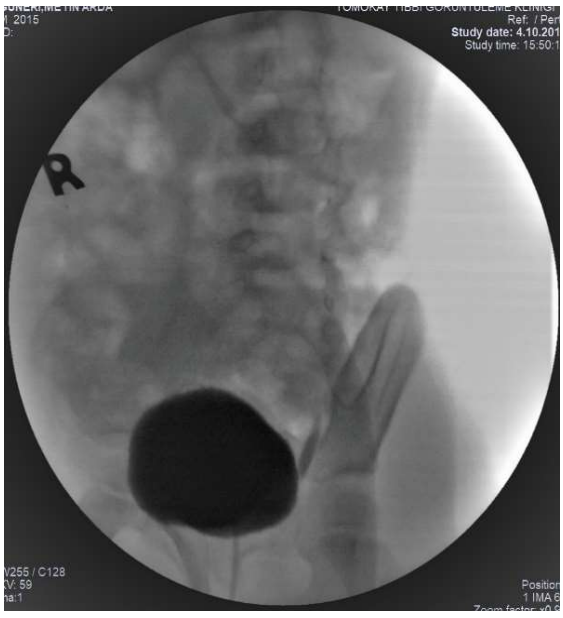

Fig. 2. Left grade 1 vesicoureteral reflux.

In order to evaluate the degree of damage in the kidney, scintigraphic evaluation showed no scar on the kidneys and the contribution of both kidneys to total renal function was equal (Sol: 49.8 Right: 50.2). We decided to perform open surgery and when bladder was opened, an edematous ureterocele was seen on the left and a single orifice on the same side (Fig. 4). A small incision was made to the ureterocele and seven pieces of stones of which the largest one was approximately seven millimeters in size, were extracted (Fig. 5, 6).

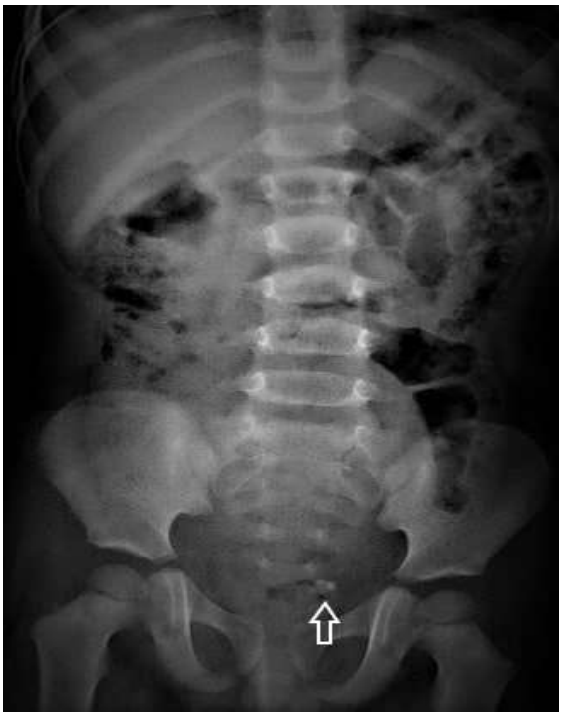

Fig. 3. The kidney-ureter-bladder radiography showed several clustered opacities.

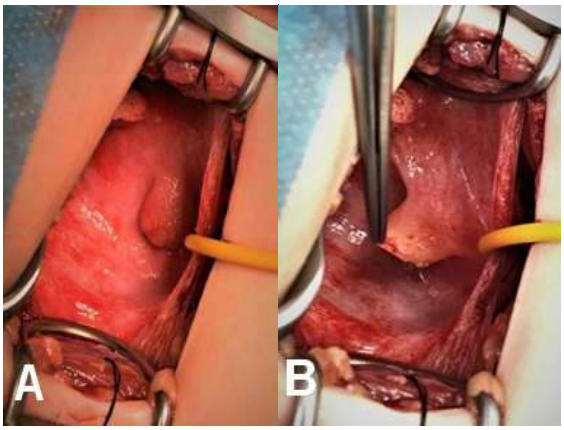

Fig. 4. Edematous ureterocele was seen on the left and a single orifice.

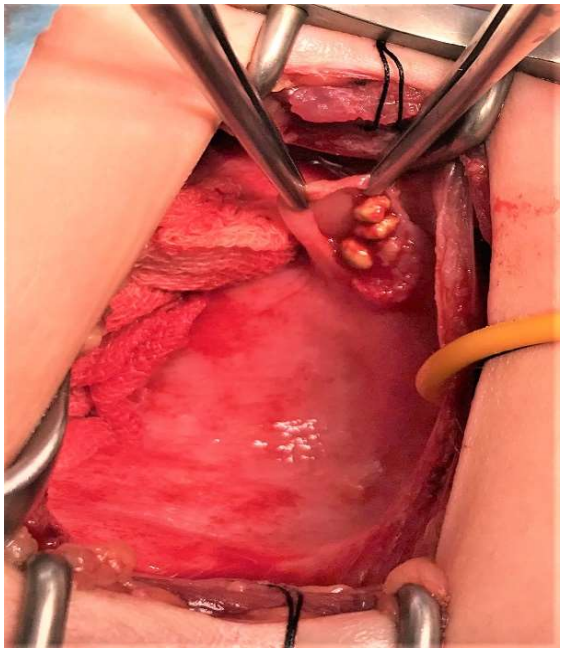

Fig. 5. Multiple stones inside ureterocele.

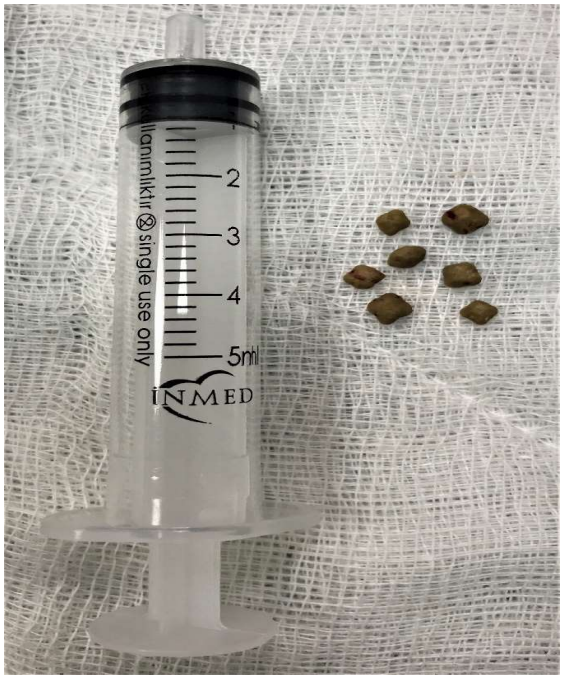

Fig. 6. View of the seven stones extracted from the ureterocele.

Retrograde pyelography (RGP) was performed by administering opaque material through the ureteral catheter which was placed in the left ureter. There was no evidence of dilatation or obstruction in the ureter. It was dissected and released by catheterization of the ureter. The released left ureter was reimplanted with the Cohen technique.

Discussion

The ureterocele is an abnormal cystic dilatation of the distal intramural tip of the ureter into the bladder [6]. The most common theory accepted for the pathogenesis is the incomplete resolution of Chwalla's membrane and consequently the development of ureteral obstruction [7,8]. Ureteroceles can lead to complications such as UTI, renal scarring and nonfunctioning upper pole, especially in duplex systems [9]. Approximately 10% of the ureteroceles are bilateral and it is most commonly seen in women with a 4: 1 ratio [8]. Although the diagnosis is made in the antenatal period or in the pediatric age with the developing technology, diagnosis may delay as late as adulthood [2]. While ureteroceles can be diagnosed up to 70% with intravenous urography and US, VCUG should be performed if necessary [9,10]. If the distal ureter is larger than the normal diameter or if a collection is seen, ureterocele should be suspected. As seen in the literature, large distal ureteral stones may mask ureteroceles [1].The detection of stones in the ureterocele is closely related to the experience of the radiologist who performed the US [9]. In our case, the ultrasound examination showed that the distal ureter stone was in the ureterocele with a clustered form.

An ureterocele may be asymptomatic or may be presented with recurrent UTI, renal dysfunction, pyelonephritis, symptoms such as renal colic (due to stone obstruction), pollakiuria, acute retention of urine, hematuria and fever. It can be presented with nonspecific abdominal or flank pain according to the anatomical location of the ureterocele [2]. In

our case, patient had symptoms of hematuria and recurrent UTI.

Dilatation in the ureterocele due to obstruction may lead to the formation of ureteral stones depending on the stasis. Other risk factors for stone formation in ureterocele are; ureteral atonia, family history and previous UTI [9]. According to the literature, stone formation in ureterocele is a rare condition in the pediatric age group with an incidence of 4% to 39% [4]. Multiple stone in ureterocele is even rarer in children [9]. There were seven stones in ureterocele in our patient and it can be valuable in this aspect. Renal colic may develop when the stone causes complete obstruction in the ureter lumen. [1]. In present case, there was no obstruction in the urine flow so the patient has no renal colic pain.

The aims of ureterocele treatment are to prevent development of VUR, UTI and loss of renal function [9]. Treatment is not required in asymptomatic patients [1]. Although the endoscopic method for ureterocele treatment is gaining popularity in the literature, stone extraction with open surgical technique can be performed in complex cases [1,10].

Endoscopic ureterocele incision is a minimally invasive method with the potential developing VUR and vesicoureteral junction obstruction after the procedure [2,11]. The fact that children are more sensitive to VUR makes the endoscopic intervention controversial in the pediatric age group [10,11,12]. Gilbert et al. reported that 8-month-old case, grade 3 VUR was developed after an endoscopic incision of ureterocele (without VUR) due to stone [13]. Gotoh et al. reported that ureteroceles treated with endoscopic incision have high rates of reoperation with open surgery between 7-25% [14].

The coexistence of single system ureterocele with stone and VUR is very rare in the literature. In present case, grade 1 VUR was present and an endoscopic intervention was not performed with the concern of increase in existing reflux level. Suprapubic cystostomy is proposed for stone removal in some reports, but this is a very invasive procedure for children. Because of large size of the stone (18x15x12 mm), we could not afford the risk of prolonged anesthesia and operation time with the endoscopic stone fragmentation. And the urethral removal of the fragmented parts (risk of developing urethral stricture as a result of urethral recurrent access in a 3-year-old boy) can be so problematic in pediatric patient so we decided to perform an open surgical procedure.

Single caliceal system, bilateral and orthotopic ureterocele types are less common [2]. Our case was intravesical and a single system ureterocele. In open surgery, the bladder was checked and a single orifice was seen on the left side. In this case, the filling of all calyces of the left kidney in intraoperative RGP supports our patient as a single system.

In open surgery, edematous ureterocele was incised and seven stones were removed. Gilbert et al. reported that removal of three stones from the left single system ureterocele. There are also cases in the literature that reported removal of a single stone from the ureterocele [10,13]. In present case, Cohen method was preferred because of Grade 1 VUR and high success rates in ureteral reimplantation.

Conclusion

It should be noted that ureteral stones might present in the ureterocele, especially the distal ureteral stones in which stone size exceeds normal ureter diameter. In the case of multiple stones in the ureterocele, open surgical intervention may be a good option for

treatment with a low probability of need for reoperation.

Compliance with ethical statements

Conflicts of Interest: None.

Financial disclosure: None.

Consent: All photos were taken with parental consent.

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