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Case Report

Double collecting system with ectopic ureterocele masquerading as an ovarian torsion

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ABSTRACT

Objective: Ureterocele in a duplex system is rare and commonly presented with urinary tract infection at neonatal age, infant or childhood. Symptomatic ureterocele in reproductive-age is a diagnostic challenge and should be highly awarded to avoid miss-diagnosis.
Case report: An adolescent girl with right ectopic ureterocele presented as acute abdomen that mimicked ovarian torsion received emergent laparoscopic surgery. Right ureterocele was identified and excised. Computed tomography later showed bilateral renal duplications with visible renal parenchyma and upper ureters. Recurrent abdominal pain with pelvic abscess occurred 10 days after surgery. Laparoscopic right partial nephrectomy of the upper moiety and resection of the residual ureterocele was performed. Cystoscopy showed absence of intravesical ureterocele and her symptoms were completely resolved after surgery.
Conclusion: Infected ureterocele in a duplex system is a rare condition and should be kept in mind as differential diagnosis.

Introduction

Duplication of the ureters is the most frequent malformation of the urinary tract in young ages, and females were reported to be affected more often than males [1,2]. In females, complete duplex systems had a higher prevalence of vesicoureteral reflux (VUR) and associated anomalies [1], such as obstructed ectopic ureter or ureterocele [3].

Ureterocele is defined as a congenital pseudocystic dilation of the intravesical ureter that results from a malformation of the submucosa of the bladder [4]. The incidence of ureteroceles has been reported to be around 1 in 500 to 1 in 12,000 by autopsy studies [5], suggesting that ureteroceles are not as uncommon as expected and are usually asymptomatic. Symptomatic ureterocele commonly presented as febrile urinary tract infection that occurred shortly after birth [5]. The mean age at presentation was 1.1 months (range 1–3) in a study that included 46 children with ectopic ureterocele in a duplex system that underwent primary endoscopic incision [4]. Symptomatic ureterocele presented in adulthood are usually difficult to detect and are likely to be associated with secondary complications.

Case report

This is a case of a 15-year-old nulli-gravida girl who presented to the emergency department with acute right lower abdominal pain for 1 day. She denied having sexual experience. Her menstruation period was regular, in moderate amount and without dysmenorrhea. Her last menstruation period was 9 days before her visit to emergency department. She denied having any systemic disease. On arrival, her abdominal pain was found progressing and accompanied with vomiting. Physical examination showed tenderness at right lower quadrant of abdomen without rebounding pain, muscle guarding or rigidity. She was febrile, documented as 38.9 °C, without chillness on arrival. Laboratory study showed hemoglobin of 11.4 g/dL, white cell count of 11.26 k/uL, and platelets of 209 k/uL. Her Creatinine level was 0.6 mg/dL and C-reactive protein level was 0.03 mg/dL. Urinalysis showed hematuria (20–35 red blood cells/high power field) without pyuria. Abdominal ultrasonography revealed a 7.7 cm × 5.3 cm lobulated cyst at right adnexa with homogenous echo-lucent content (Fig. 1).

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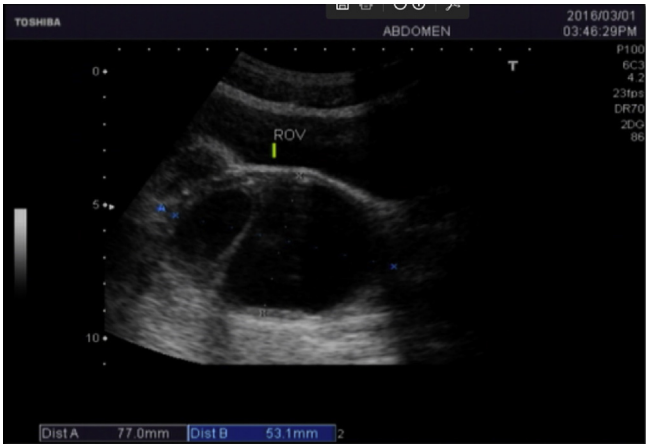

Fig. 1. Ultrasonography showed a 7.7 cm × 5.3 cm lobulated cyst with clear echolucent content at right adnexa.

Emergent single incision laparoscopic surgery was performed and identified a 9 cm × 2 cm right retroperitoneal tubal cyst with turbid fluid content. The distal and caudal ends of the tubal cyst extended from the obturator fossa to upper retro-infundibular space (Fig. 2). After identification of her right ureter, the tubal cyst was excised and a retroperitoneal drainage tube was inserted. The two ends of the tubal cyst were un-ligated. The nature of the tubal cyst was unknown during surgery. Due to the emergency condition, other surgical specialist such as Urologist was not consulted. Antibiotics were given and she was discharged uneventfully after drainage tube removal 6 days after operation. Pathology examination revealed a uroepithelial lining cyst, suggesting of a ureterocele (Fig. 3).

Computed tomography after operation showed bilateral renal duplications with visible renal parenchyma and upper ureters. The upper moiety of the right kidney that connected to the ureterocele was small, and conservative treatment was suggested. Unfortunately, she suffered from abdominal pain 10 days after operation. Subsequent computed tomography showed intact right kidney, dilatation of right upper ureter and localized pelvic abscess (Fig. 4), suggested possible stricture at the lower segment of right upper moiety ureter. Laparoscopic partial nephrectomy of right upper moiety and resection of the residual ureterocele were performed. Dilated right upper moiety and ureterocele with dark-brownish turbid content was found. Cystoscopy revealed one right ureteral orifice and two left ureteral orifices. Neither intravesical ureterocele in the bladder nor ectopic ureteral orifice in the vagina was seen. She recovered uneventfully after surgery. Track back to her history, she reported to have frequent vaginal watery dripping since birth without definite diagnosis. This symptom was completely resolved after the two surgeries.

Discussion

Acute pelvic pain is a common cause of emergency department visits. Differential diagnosis of acute abdomen in women includes gynecologic etiologies, such as pelvic inflammatory disease, ruptured ectopic pregnancy, and ovarian torsion; and non-gynecologic causes such as appendicitis, pancreatitis, ischemic colitis, bowel perforation, cystitis, and acute pyelonephritis [6,7]. The conditions range from a self-limiting disease to a surgical emergency. In this case, an adolescent who presented to the emergency department with acute pelvic pain was initially diagnosed as ovarian torsion based on her acute symptoms and laboratory tests. She was later diagnosed as infected right ectopic ureterocele in a duplex system, which is a rather rare symptomatic disease in her age.

Ultrasonography is a tool available in the Emergency Department for the immediate diagnosis of acute abdomen. A unilateral enlarged ovary could be found in 71.3% of acute abdomen in pediatrics, and presented as complex (53.6%), cystic (27%), solid (15.1%), or calcified (2.4%) in sonographic images [5]. The appearance of ureterocele and the dilated upper urinary tract in ultrasonography varied widely [4] and differential diagnosis is difficult.

In pediatrics, 75% of ureteroceles was reported in associated with a duplex urinary system [8]. The caudal end of the ureterocele typically occurs in the ureter that drains the upper pole of the kidney [3]. The distal ends of ureteroceles were classified as intravesical or extravesical type according to its related position to the bladder and/or the presence of intrinsic obstruction of the ureteral orifice [9]. In our patient, the distal opening of her ureterocele was not identified in the bladder. It is likely that her ectopic ureterocele was an extravesical type and the opening of ureterocele

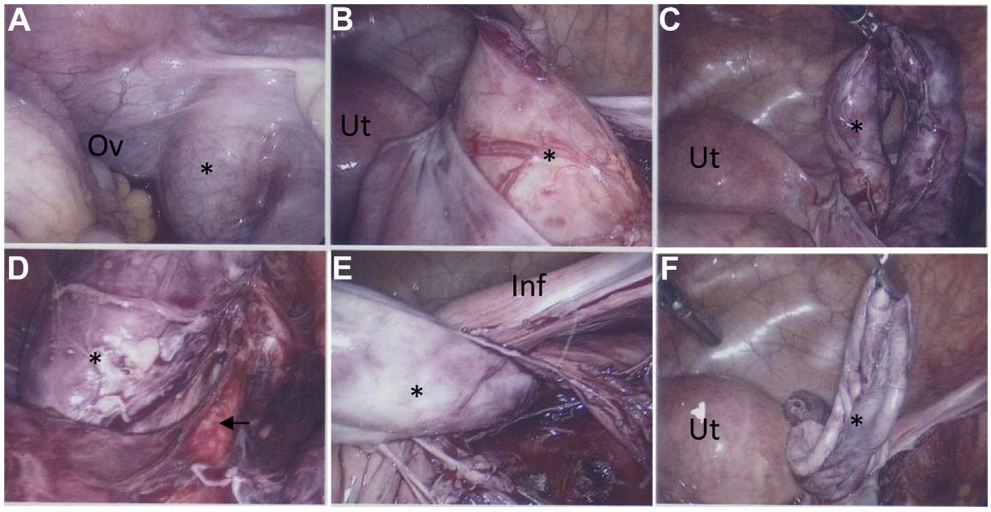

Fig. 2. Intraoperative photography of the endoscopic procedure. (A) A retroperitoneal bulging cystic structure was identified (*). (B) The cyst structure was dissected from the retroperitoneal space. (C) A 9 cm × 2 cm retroperitoneal tubal cyst was dissected with distal end extending into the obturator fossa. (D) The right ureter (arrow) was well identified. (E) The caudal end of the tubal cyst extended upward below infundibular ligament. (F) The tubal cyst with turbid fluid content was excised and removed. Inf = infundibular ligament; Ov = ovary; Ut = uterus.

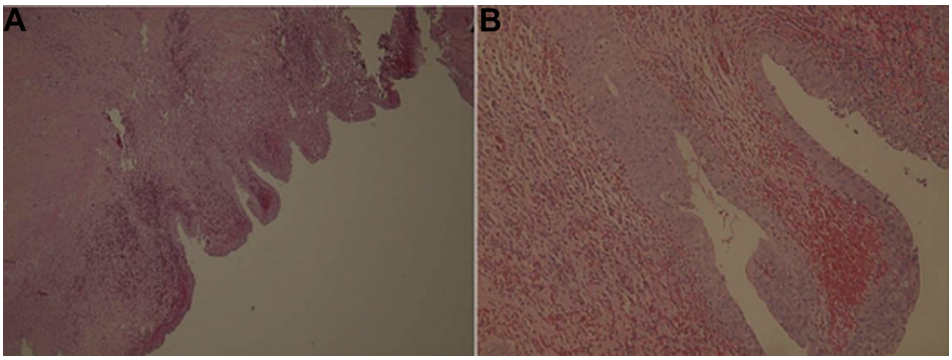

Fig. 3. Histopathology showed a well-organized tubal structure with urothelial-like lining and muscular wall (A: magnification 4X) and foci of inflammation and hemorrhage (B: magnification 10X). (Hematoxylin & Eosin stain.)

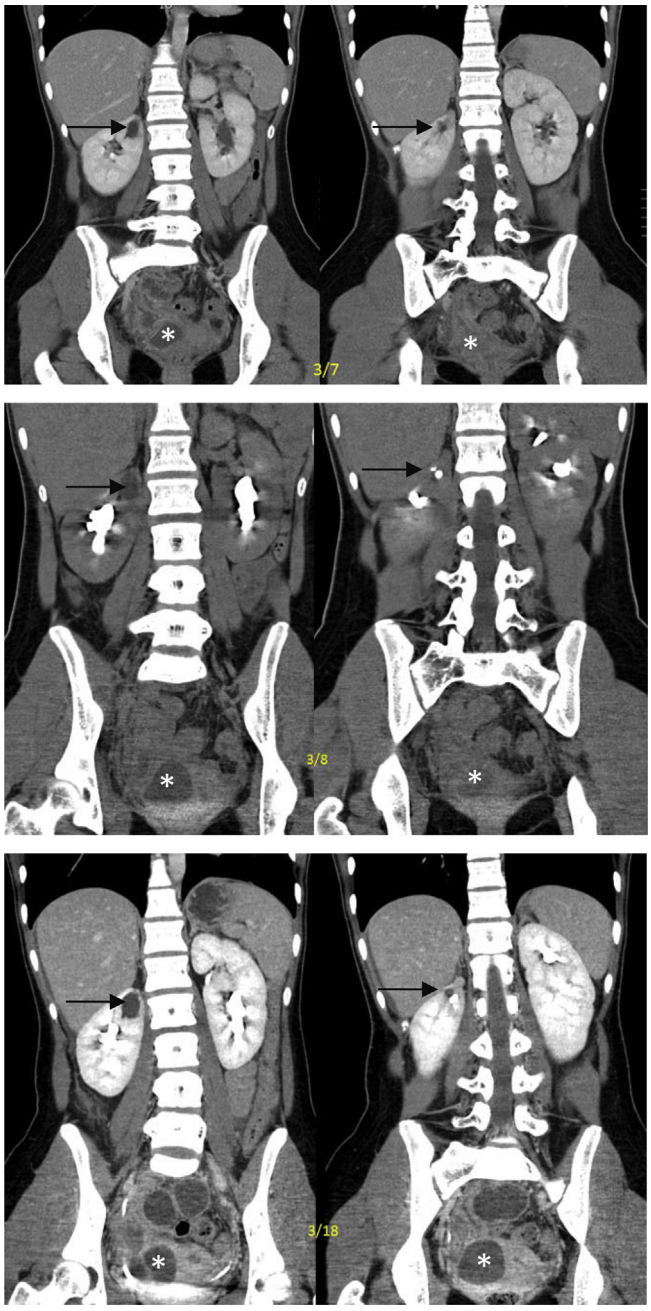

Fig. 4. Serial computed tomography scans after first surgery showed renal duplications with visible renal parenchyma and upper ureters. The right upper moiety ureter was dilated (arrow) and localized abscess (*) was seen in the right pelvis.

was in the vagina that caused her watery (urine) drippling since birth. However, identification of extravesical opening could be difficult after excision of ureterocele.

The goals of management in ureterocele includes prevention of renal damage associated with obstruction or VUR and urinary tract infection, promotion of continence, and preserve the function of renal moiety [10]. However, the means of accomplishing these objectives remain a significant challenge [11]. Close observation without surgical management has been advocated in patients with ureterocele that were presented as non-obstructed ipsilateral inferior moieties, absence of bladder outlet obstruction, nonfunctioning superior moieties, and less than high grade VUR [12]. Some pediatric surgeons prefer endoscopic ureterocele decompression or puncture for children and neonates with small, asymptomatic, or with low-grade reflux in nonobstructive intravesical ureteroceles or extravesical ureteroceles [11].

In ectopic ureterocele, or duplex system and high-grade preoperative VUR, it was reported that ureterocele excision could only resolve about 50% of VUR [13]. Additional secondary operative procedures such as excision of the non-functioning upper moiety were often required to treat iatrogenic VUR [2,11]. Therefore, to have a better resolution for symptomatic duplex system, two types of surgical options, the lower and the upper tract approaches were suggested. In lower tract approach, ipsilateral ureteroureterostomy (IUU) is an increasingly used alternative for children with duplication in whom the obstructed moiety has significant functionality [11]. The upper tract approach included heminephrectomy with partial ureterectomy has been advocated as the primary treatment of duplex system [13]. However, the disadvantage of heminephrectomy was vascular compromise and associated loss of lower-moiety function [14]. In our patient, ureteroureterostomy was not performed due to the reasons that ureterocele was not identified during the emergent operation, it is an infected situation, and the condition of the distal end of ureterocele was unknown.

Some surgeons advocated more aggressive approach, single-stage reconstruction that included superior moiety (SM) heminephrectomy, ureterocele excision, bladder base/neck reconstruction, and inferior moiety (IM) ureteral reimplantation at the bladder level during initial surgery [2,11]. However such procedures are dangerous and carry a risk of damaging the bladder innervation [2,11]. In contrast, conservative approach by leaving behind a nonfunctioning or poorly functioning renal moieties in situ after primary excision of ureteroceles is not well understood. The risk of possible morbidity (eg, infection, hypertension, malignancy) or necessitate subsequent heminephrectomy remain unknown.

In our case, the upper moiety and the residual ureterocele showed persisting infection after primary excision of ureterocele that causes subsequent secondary operation unavoidable. Track back to her history, if ureterocele was suspected before surgery,

complete image work-up, cystoscopic examination, combined with antibiotics treatment could have been given followed by a complete laparoscopic resection of ureterocele and partial nephrectomy of right upper moiety in one stage operation. On the other hand, if incomplete resection of ureterocele was noticed during surgery, urologist could have been consulted to perform a wider exploration of surgical field to perform a more complete surgery. However, due to the rare incidence of symptomatic ureterocele presented in adulthood, she was delayed diagnosed in the emergency situation. Fortunately, her symptoms relieved completely after the two surgeries.

Conclusion

The anatomical and clinical characteristics of ureterocele in a duplex system vary widely. An adolescent with an infected ureterocele could be misdiagnosed as ovarian torsion. Cautious should be paid in the differential diagnosis of patients with acute pelvic pain and history of abnormal vaginal discharge.

Consent

Written informed consent was obtained from the patient for publication of this case report and the accompanying image and use of the images.

Authors' contribution

CY Wu: Data collection, Data analysis, Manuscript writing.
CY Lee: Data analysis; Manuscript editing.
IJ Yang: Data analysis; Manuscript editing.
H Shen: Data analysis; Manuscript editing.
PL Torng: Protocol/Project development, Data analysis, Manuscript editing.

Financial disclaimer/Conflict of interest

None.

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