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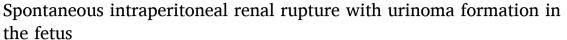
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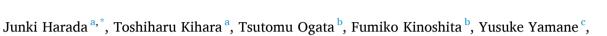
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ABSTRACT

Spontaneous intraperitoneal renal rupture with urinoma formation in fetuses is an unusual condition that is caused by upper or lower urinary tract obstruction. We report the case of a neonatal male infant who presented with a spontaneous intraperitoneal right renal rupture accompanying ipsilateral ureterovesical junction obstruction (UVJO). Fetuses with UVJO accompanying contralateral multicystic dysplastic kidney should be observed carefully because of the risk of spontaneous renal rupture.

Introduction

Spontaneous intraperitoneal renal rupture with urinoma formation is a rare urological condition that is caused by upper or lower urinary tract obstruction. Additionally, a recent research indicated that urinoma may be related to poor fetal renal function. Herein, we report a rare case of spontaneous intraperitoneal renal rupture with urinoma formation in a fetus caused by ipsilateral ureterovesical junction obstruction (UVJO).

Case presentation

A neonatal male infant was referred to our department immediately after an emergency caesarean section at 34 weeks' gestational age (GA) for severe right hydronephrosis with perirenal fluid collection, left multicystic dysplastic kidney (MCDK), ascites, and pleural effusion. The 28-year-old mother arrived at the department of obstetrics and gynecology of our hospital at 24 weeks' gestation for a fetal abdominal simple cystic mass of approximately 1.5-cm in diameter. Ultrasound findings showed a cystic mass of the same size near the upper left side of the urinary bladder, but there was no oligohydramnios or other abnormalities. In contrast, magnetic resonance imaging (MRI) revealed that the left kidney had a cystic mass leading to the inferior pole, and multiple small cysts (Fig. 1). MRI also revealed right ureteropelvic dilatation. Based on these findings, right UVJO and left MCDK were suspected. However, at 34 weeks' GA, ultrasound findings showed anhydramnios

and increased right hydronephrosis. MRI revealed right perirenal fluid, ascites, and pleural effusion (Fig. 2). Therefore, an emergency caesarean section was decided. Ultrasound examination of the neonatal infant also showed right perirenal fluid and no urine storage in the bladder. His serum creatinine level was 0.86 mg/dL. One possible explanation was that perirenal urinoma, which was secondary to spontaneous rupture of the right hydronephrosis, had ruptured into the peritoneum, and intraperitoneal urine leakage led to ascites, pleural effusion, and increasing serum creatinine level. Therefore, exploratory laparoscopy was performed to examine and repair the intraperitoneal renal rupture.

On exploratory laparoscopy, hemorrhagic ascites was present, and intraperitoneal rupture with continuous urine leakage near the right lateral inguinal fossa and a yellowish right ureter were identified (Fig. 3A and B). Furthermore, a 1.5-cm rupture of the right renal parenchyma with urinary leakage was observed by retroperitoneal approach. This rupture was catheterized using an 8-Fr balloon catheter and repaired using 3–0 absorbable sutures (Fig. 3C and D) with retroperitoneal drain tube insertion.

The postoperative course was uneventful, except for temporary emesis, and first urination was observed at 19 days after the operation. Antegrade pyelography performed at 10 days after the first urination depicted the right pelvis and ureter clearly, but the bladder was depicted slightly (Fig. 3E). In contrast, voiding cystourethrography did not show posterior urethral valve (Fig. 3F). The balloon catheter was removed 4 days after performing contrast radiography. The infant was discharged

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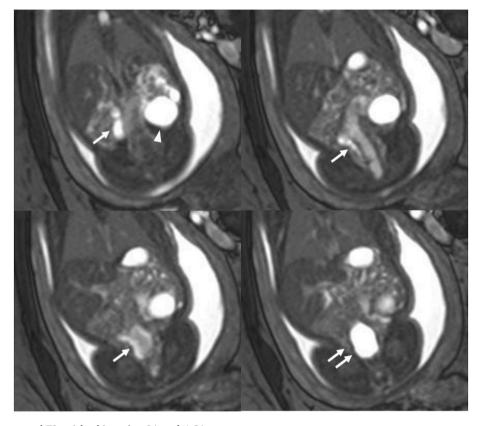


Fig. 1. MRI findings (fat-saturated T2 weighted image) at 24 weeks' GA. Single and double arrows showing right ureteropelvic dilatation with urine storage in bladder. Arrowhead showing left kidney with a 1.5-cm cystic mass and multiple small cysts.

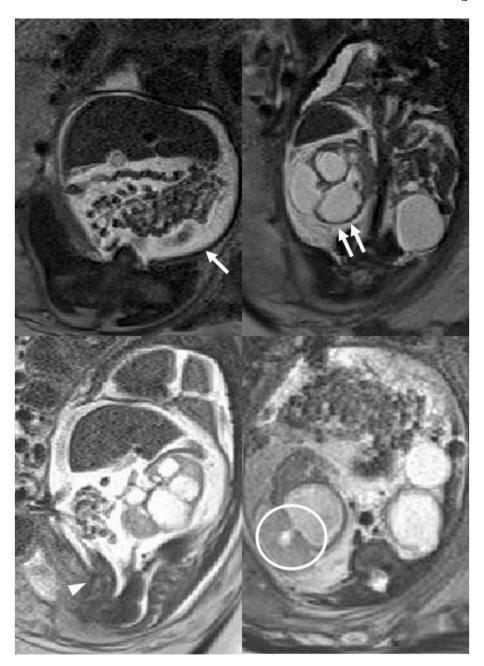


Fig. 2. MRI findings (half-Fourier acquisition single-shot turbo spin-echo) at 34 weeks' GA.
Ascites (single arrow) and enlargement of the right fetal hydronephrosis (double arrows) without urine storage in the bladder (arrow head) being observed. The circle showing a part of rupture.

48 days postoperatively, with a serum creatinine level of 0.37 mg/dL. No urologic complications were observed at the 5-month follow-up periods and the serum creatine level was 0.38 mg/dL.

Discussion

Spontaneous intraperitoneal renal rupture with urinoma formation is a rarely reported urological condition. Urinoma is an encapsulated accumulation of urine extravasation into the perirenal space, which is caused by urine leakage from the renal collecting system. Urinoma can be caused by urinary tract obstructions such as posterior urethral valves, ureteropelvic junction obstructions, neurogenic bladder, or UVJOs. Adorisio et al. reported that 62.5%, 29.5%, and 8% of urinoma cases resulted from obstructions in the lower urinary tract, obstructions in the

upper urinary tract, and from unknown causes, respectively.3

It has suggested that urinoma formation is part of the protective system of renal parenchyma called a pop-off mechanism to buffer high pressures in the urinary tract and reduce renal dysplasia, but a recent study showed that urinoma formation damages the affected kidney and is a possible prognostic factor for poor renal function. ³

In general, fetal urine production increases with GA. Lee et al. reported that after 25 weeks' gestation, the urine production rate increased rapidly every 2 weeks. The increasing urine production may lead to rapid increase in the internal pressure of the upper urinary tract. Therefore, urine extravasation and urinoma formation may more likely occur after 25 weeks' GA. One study reported that the mean GA at the time of urinoma formation was 25.7 (range, 18–36) weeks. ¹

In the present case, UVJO was considered to be the cause of urinary

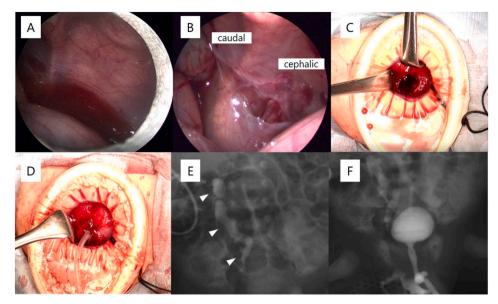


Fig. 3. Intraoperative findings, antegrade pyelography, and voiding cystourethrography after operation.

Exploratory laparoscopy (A, B) and intraoperative field (C) showing hemorrhagic ascites, intraperitoneal rupture with yellowish ureter through the peritoneum, and a 1.5-cm rupture of the right renal parenchyma. Renal parenchymal rupture repaired using 3–0 absorbable sutures with balloon catheter (D). Antegrade pyelography and voiding cystourethrography findings revealing right UVJO (arrowheads) without posterior urethral valve (E, F).

tract obstruction because the first fetal MRI and postnatal antegrade pyelography findings revealed right ureteropelvic dilatation. Additionally, increased urine production of the right kidney might have compensated for the left MCDK. The abnormal urine production increase of the right kidney could have placed stress on the right upper urinary tract with UVJO, leading to renal parenchymal rupture and urinoma formation.

Conclusion

We described an unusual case of spontaneous fetal renal rupture into the peritoneum with urinoma formation. Fetuses with UVJO, especially those with accompanying contralateral MCDK, should be examined carefully on imaging because of the risk of spontaneous renal rupture.

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Declarations of competing interest

None.

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