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Page kidney in a child with ureteropelvic junction obstruction of lower moiety in a partial duplex system

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ABSTRACT

Reports of hydronephrosis-induced hypertension in pediatric patients are rare. A 7-year-old girl with hypertension was referred to our hospital with left hydronephrosis caused by ureteropelvic junction obstruction of the lower moiety in a partial duplex system. Because the lower moiety was almost nonfunctional, a laparoscopic heminephrectomy was performed. Antihypertensive drugs were unnecessary on the day after surgery. This is the first report of a Page kidney in a patient with ureteropelvic junction obstruction of the lower moiety. The cause of hypertension was evaluated based on the plasma renin activity and pathological findings of the lower moiety.

1. Introduction

Page kidney (PK) is a rare condition that presents with hypertension (HT) secondary to compression of the renal parenchyma.[1,2] Most cases of PK have been reported in adults following trauma,[2] and nontraumatic PK in children is extremely rare.[3] Moreover, PK typically arises from external compression of the renal parenchyma.[1,2] However, we propose that both external compression can cause PK but also internal-to-external compression of the renal parenchyma due to severe hydronephrosis.

Herein, we report a case of PK in a child with ureteropelvic junction obstruction (UPJO) of the lower moiety in a partial duplex system that was diagnosed based on postoperative normalization of blood pressure and plasma renin activity (PRA) after laparoscopic heminephrectomy (LHN) for hydronephrosis of the lower moiety.

2. Case report

A 7-year-old girl with abdominal distension was admitted to a pediatric hospital. The patient had a history of immunoglobulin A vasculitis. She was diagnosed with HT and left hydronephrosis and was started on antihypertensive medication. Subsequently, the patient was referred to our hospital for further investigation of left hydronephrosis.

Abdominal computed tomography revealed a left partial duplex system with UPJO of the left lower moiety (Fig. 1A and B). Differential renal function (DRF) assessed using a dimercaptosuccinic acid scan was 31.1 % for the left kidney and 1.7 % for the left lower moiety (Fig. 1C). Given the significantly reduced function of the left lower moiety, we planned for LHN.

Initially, cystoscopy was performed under general anesthesia with the patient in the lithotomy position. Cystoscopy revealed that the right and left ureteral orifices were in normal anatomical positions. Subsequent retrograde pyelography revealed a left-sided partial duplex system. A 4-F ureteral catheter was placed in the ureter of the left upper moiety. The patient was then placed in an oblique position, with the operated side elevated. LHN was performed using the retrocolic approach. A 12-mm trocar was inserted as a camera port into the peritoneal cavity through an incision in the upper part of the umbilicus, and two 5-mm trocars were inserted as working ports. The procedure revealed a dilated pelvis in the lower moiety that significantly obstructed the operative field. Subsequently, a controlled incision was made into the dilated pelvis to aspirate renal urine, facilitating adequate visualization for further surgery (Fig. 2A). Laparoscopic exploration successfully identified the Y-shaped left ureter. Arteries and veins supplying the lower moiety were clearly visualized. Using LigaSure bipolar forceps (Medtronic PLC; Minneapolis, MN, USA), the vessels were

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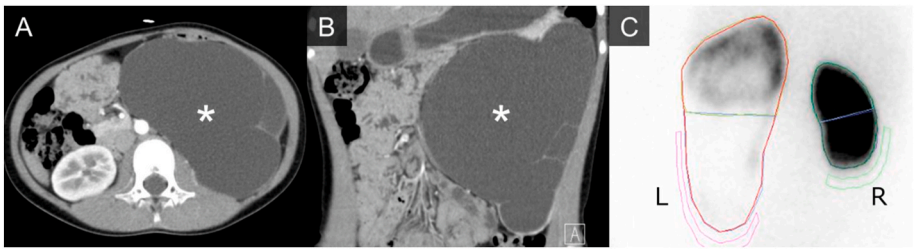

Fig. 1. Preoperative imaging examination findings. (A), (B) Contrast-enhanced computed tomography showing hydronephrosis of the lower moiety (asterisk). The renal pelvis is severely dilated, compressing the renal parenchyma of the lower moiety. (A) Transverse plane. (B) Coronal plane. (C) The dimercaptosuccinic acid scan indicates that the differential renal functions of the left kidney and left lower moiety are 31.1 % and 1.7 %, respectively.

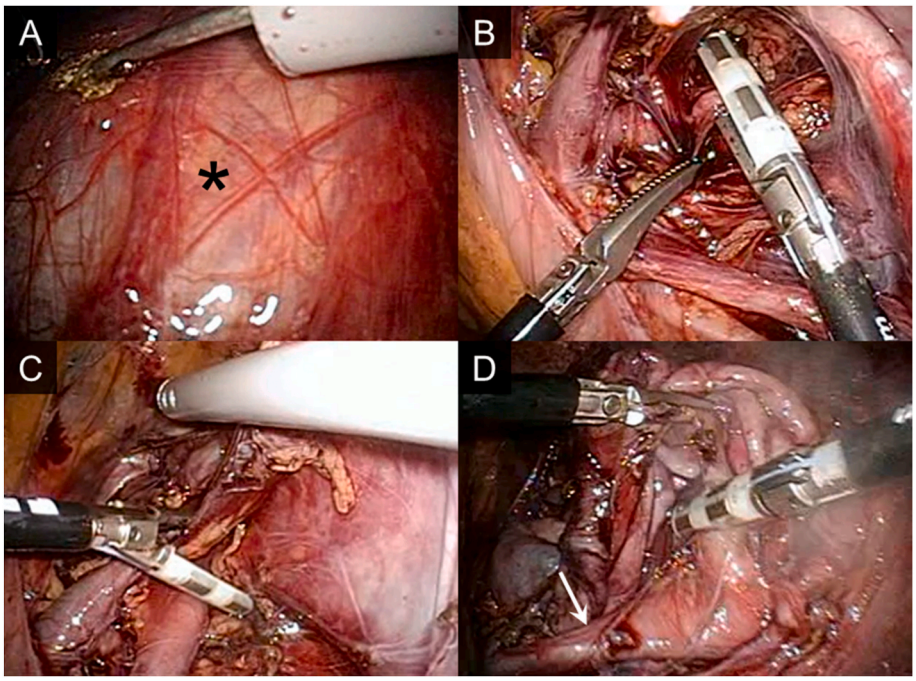

Fig. 2. Findings of laparoscopic heminephrectomy. (A) The dilated pelvis of the lower moiety (asterisk) is incised, and renal urine is aspirated to ensure the operative field. (B) The lower moiety artery is closed and cut using LigaSure bipolar forceps. (C) The lower moiety vein is closed and cut using LigaSure bipolar forceps. (D) The ureter of the lower moiety is closed and cut using LigaSure bipolar forceps. The arrow indicates the ureter of the upper moiety.

meticulously sealed and transected (Fig. 2B and C). Similarly, the ureter of the lower moiety was transected and closed using LigaSure (Fig. 2D). Subsequently, the lower moiety was dissected using a monopolar hook and removed through the umbilicus (Fig. 3A).

Antihypertensive drugs were discontinued because the patient's blood pressure normalized on the day after surgery. The levels of PRA and aldosterone after surgery also normalized to 2.8 ng/mL/hr and 93 pg/mL. However, the preoperative levels of PRA and aldosterone were high at 8.5 ng/mL/hr and 254 pg/mL, respectively. Focal glomerular structures were observed in the renal parenchyma of the resected tissues (Fig. 3B). Immunostaining with an anti-renin antibody (ab212197; Abcam, Cambridge, England) showed that renin was intensely and focally expressed in the juxtaglomerular (JG) apparatus (Fig. 3C and D). Based on the normalization of blood pressure and PRA after LHN, the patient was diagnosed with HT due to PK disease. Ten years have passed since the operation, the DRF of the left upper moiety has been preserved, and the patient's blood pressure has remained normal.

3. Discussion

The pediatric nephrological literature identifies UPJO as a cause of HT.[4] However, HT caused by UPJO is rare,[5,6] although patients with UPJO may develop HT later.[7] Although the pathogenesis of HT in patients with UPJO is unclear, hyperreninemia may be a cause thereof.[8] Moreover, it has been reported that hypertrophy of the JG apparatus is observed in the renal parenchyma,[8,9] and renin is strongly expressed in the JG apparatus by pathological examination of patients with UPJO complicated by HT.[9]

PK is a cause of HT and is typically associated with trauma in adult patients.[1,2] PK is diagnosed by imaging findings that show that subcapsular contents, such as hemorrhage or urine, compress the renal parenchyma.[2] Recently, rare nontraumatic cases in pediatric patients have been reported.[3] Although the pathogenesis of PK is also unclear, one of the causes is thought to be the activation of the renin–angiotensin system due to ischemia of the renal parenchyma by the compression of the renal parenchyma.[10] In addition to imaging findings, there are also reports of PK showing an increase in PRA.[11] Furthermore, an adult case of PK due to intraperitoneal compression of the kidney has been reported.[12] This suggests that PK can also develop by compression of the renal parenchyma from contents other than subcapsular.

The patient was diagnosed with HT because of PK for the following reasons: First, the blood pressure of the patient normalized immediately after LHN, although preoperative imaging findings showed that the severely dilated renal pelvis compressed the renal parenchyma of the

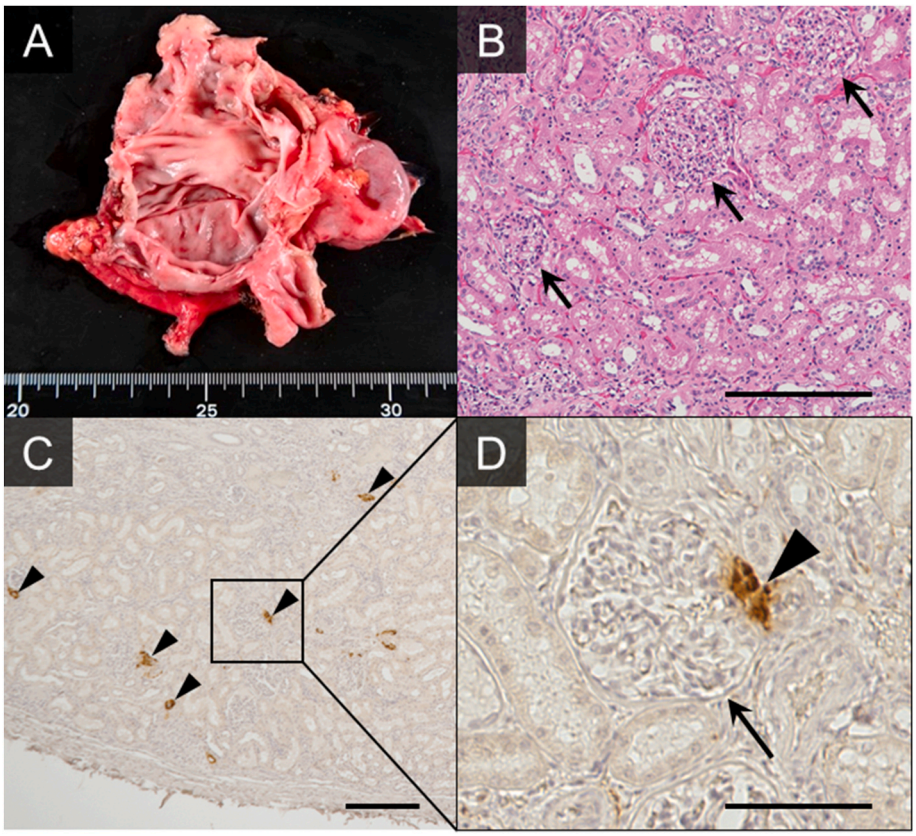

Fig. 3. The excised tissue (A) and pathological findings (B–D). (A) The pelvis of the lower moiety is sac-shaped, and the thin renal parenchyma partially remains. (B) Hematoxylin and eosin staining shows that the glomeruli (arrows) remain focally in the renal parenchyma. The scale bar indicates 250 μm. (C), (D) Immunostaining with anti-renin antibody. (C) Low-power field. Renin focally expresses intensely in the juxtaglomerular (JG) apparatus (arrowheads) of some glomeruli. The scale bar indicates 250 μm. (D) High-power field. Hypertrophy of the JG apparatus is observed, and renin is extensively expressed in the JG apparatus (arrowhead). The arrow indicates the glomerulus. The scale bar indicates 100 μm.

lower moiety. Second, the PRA level normalized after LHN, although the level of PRA was high before LHN. Therefore, we concluded that HT was due to hyperreninemia caused by the compression of the renal parenchyma. This is the first report of PK demonstrating high renin expression in the JG apparatus by pathological evaluation.

PK is usually diagnosed as a result of trauma or some events.[1–3,10–12] In other words, patients with PK seem to be in a situation where it is easy to check the presence or absence of HT by checking their vital signs. However, the patient in this case had not been diagnosed with HT until the patient was admitted to the referring hospital because the patient solely presented with abdominal distension. Therefore, the timing of HT onset in this patient was unclear. However, the pathogenesis of HT in some patients with UPJO may be similar to that of PK if HT in both patients with PK and patients with UPJO is caused by hyperreninemia.[8–11]

4. Conclusion

We successfully diagnosed PK based on the normalization of blood pressure and PRA after LHN. The possibility of PK should be considered when HT complicates severe hydronephrosis because HT is extremely rare in pediatric patients. Prompt surgical intervention for hydronephrosis complicated by HT may protect renal function.

CRediT authorship contribution statement

Hidenori Nishio: Writing – original draft, Data curation, Conceptualization. **Kentaro Mizuno:** Writing – review & editing. **Takuya Sakata:** Data curation. **Daisuke Matsumoto:** Data curation. **Hideyuki Kamisawa:** Writing – review & editing. **Satoshi Kurokawa:** Writing – review & editing. **Akihiro Nakane:** Writing – review & editing. **Tetsuji Maruyama:** Supervision. **Takahiro Yasui:** Supervision. **Yutaro Hayashi:** Supervision.

Financial disclosure

None.

Declaration of competing interest

None.

References

1. Engel WJ, Page IH. Hypertension due to renal compression resulting from subcapsular hematoma. *J Urol.* 1955;73:735–739. https://doi.org/10.1016/S0022-5347(17)67466-4.
2. Haydar A, Bakri RS, Prime M, Goldsmith DJ. Page kidney—a review of the literature. *J Nephrol.* 2003;16:329–333.
3. Kamaraj SG, Kochunny A, Kathirvelu G, Singh P, Samnakay N. Page kidney in a child with severe pelviureteric junction obstruction. *Case Rep Urol.* 2020;2020, 8883546. https://doi.org/10.1155/2020/8883546.
4. de Waard D, Dik P, Lilien MR, Kok ET, de Jong TP. Hypertension is an indication for surgery in children with ureteropelvic junction obstruction. *J Urol.* 2008;179:1976–1978. https://doi.org/10.1016/j.juro.2008.01.058. ; discussion 1978–1979.
5. Farnham SB, Adams MC, Brock JW 3rd, Pope 4th JC. Pediatric urological causes of hypertension. *J Urol.* 2005;173:697–704. https://doi.org/10.1097/01.ju.0000153713.46735.98.
6. Bilge I. Symptomatology and clinic of hydronephrosis associated with uretero pelvic junction anomalies. *Front Pediatr.* 2020;8:520. https://doi.org/10.3389/fped.2020.00520.

7. Lee HE, Park K, Choi H. An analysis of long-term occurrence of renal complications following pediatric pyeloplasty. *J Pediatr Urol*. 2014;10:1083–1088. https://doi.org/10.1016/j.jpurol.2014.03.015.
8. Kawano S, Yano S, Takahashi S, Nomura Y, Ogata J. A case of hypertension due to unilateral hydronephrosis in a child. *Eur Urol*. 1986;12:357–359. https://doi.org/10.1159/000472656.
9. Mizuiri S, Amagasaki Y, Hosaka H, et al. Hypertension in unilateral atrophic kidney secondary to ureteropelvic junction obstruction. *Nephron*. 1992;61:217–219. https://doi.org/10.1159/000186876.
10. Dopson SJ, Jayakumar S, Velez JC. Page kidney as a rare cause of hypertension: case report and review of the literature. *Am J Kidney Dis*. 2009;54:334–339. https://doi.org/10.1053/j.ajkd.2008.11.014.
11. Duchene DA, Williams RD, Winfield HN. Laparoscopic management of bilateral page kidneys. *Urology*. 2007;69:1208.e1–1208.e3. https://doi.org/10.1016/j.urology.2007.02.040.
12. Galos P, Hultström M, Creusen AD, Eggertsen G, Urdzik J, von Seth M. Case report: an unusual presentation of renal hypertension after damage control surgery. *Int J Surg Case Rep*. 2021;82, 105872. https://doi.org/10.1016/j.ijscr.2021.105872.