Case report: Ureteral Fibroepithelial Polyps causing ureteropelvic junction obstruction in a child with a horseshoe kidney

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Case Report

Keywords: Ureteral Fibroepithelial Polyps, Horseshoe Kidney, Pelviureteral Junction Obstruction, Sepsis, Case report

Posted Date: April 15th, 2022

DOI: https://doi.org/10.21203/rs.3.rs-1526425/v1

Abstract

Background

Ureteral Fibroepithelial Polyps (UFPs) are benign tumors that arise from the mesodermal layer. The disease is a rare cause of **ureteropelvic** junction obstruction (UPJO), particularly in children.

Case presentation

we present a case of a child preoperatively diagnosed with UPJO and urogenic sepsis owing to horseshoe kidney using magnetic resonance imaging(MRI). During surgery, we found that the ureteral polyp rather than the horseshoe kidney was the cause of the PUJO.Thenus, the ureteral polyp was resected, and end-to-end anastomosis was performed with the renal pelvis and ureters. An F5 double J tube was indwelling and removed by ureteroscopy three months later. No special adverse events were found during 1-year follow-up.

Conclusions

When the clinical and radiographic findings of the patient are incompatible with the common etiology, ureteral fibroepithelial polyps should be considered as a rare cause of PUJO.

Background

Etiologies of ureteropelvic junction obstruction (UPJO) comprise primary and secondary obstruction. However, the most common cause of UPJ obstruction is congenitally narrowed pelviureteral junction.[1] Ureteral Fibroepithelial Polyps (UFPs) are a rare primary cause of **ureteropelvic** junction obstruction (UPJO), particularly in children. UFPs are benign tumors from the mesodermal layer, consisting of a fibrous vascular core, which arecovered by normal urothelial epithelium. They are mostly found in the ureteropelvic junction or upper ureter, more common among boys than girls. UFPs can grow up to 17cm.[2,3] Symptoms associated with UFPs include hematuria, flank pain, or urinary tract infection. [1–3] Horseshoe kidney is the most common renal fusion anomaly that can cause urinary obstruction. The horseshoe kidney often has abnormal kidney rotation, making the renal pelvis be in the forward position, renal calices be in the backward position, ureteral crossing across the renal isthmus which is fused by the kidneys at the lower poles and causes hydronephrosis.[4]

This study presents a case of a child preoperatively diagnosed with UPJO and urogenic sepsis owing to horseshoe kidney based on the outcomes of the clinical presentation and magnetic resonance imaging(MRI). During surgery, we found that UFPs were the primary cause of the **ureteropelvic** junction obstruction(UPJO).

Case Presentation

A 3-year-old girl with recurrent fever was admitted to the hospital for one week and diagnosed with hydronephrosis on the right kidney. On admission, the girl's temperature was up to 38.5°C, accompanied by frequent urination and dysuria. In addition, she had a history of allergy to penicillin and cephalosporin antibiotics.

Admission diagnosis: 1. Acute urinary tract infection 2. Right pelviureteral junction obstruction with hydronephrosis. Clindamycin anti-inflammatory treatment was administered after admission. On the second day, a sudden high fever occurred, reaching 40.7℃, which was accompanied by chills. The patient's white blood cell count was elevated at 20×109/L, and her serum creatinine level was normal. The imipenem 0.5 g bid was administered after urgent blood culture, and the temperature returned to normal after 3-day anti-infection treatment. The blood and urine cultures were examined to detect Escherichia coli, and the patient was discharged after a stable condition. A week later, she was readmitted to the hospital for fever with a temperature rise to 40℃, and a right renal percutaneous nephrostomy was performed. The puncture fluid culture was performed to detect Escherichia coli again. After the patient's temperature was controlled, a surgery was performed at a selected time.

Magnetic Resonance Urography (MRU) examination shows that the right kidney is in full shape; the right renal pelvis and calyces are dilated, and the lower kidneys are joined at the midline. Obstruction of the ureteropelvic junction was the cause of severe hydronephrosis of the right kidney (Fig. 1A,B). Anterograde pyelography was performed to determine the location and length of the stenosis preoperatively. The right renal pelvis and calyces were dilated, No ureteral stenosis was found because the nephrostomy tube was passed through the ureteropelvic junction (Fig. 1C,D). Thus, the incomplete obstruction of the right upper ureteral is considered.

Right pyeloplasty was performed under general anesthesia after the infection was controlled. During the operation, no obvious stenosis was observed. Thus, we opened the ureteropelvic junction and found a 2 cm polyp that was blocking the ureter. The ureteral polyp was resected, end-to-end anastomosis was performed with the renal pelvis and ureters, and an indwelling F5 double J tube was inserted. The postoperative pathological examination confirmed a benign fibroepithelial polyp (Fig. 2). The F5 double J tube was removed by ureteroscopy three months later. No special adverse events were found during 1-year follow-up.

Discussion And Conclusions

In children, UFPs are a rare primary cause of UPJO and more common in boys than girls. UFPs commonly appear on the left side, and the diagnosis rate is 22% before operative.[5] They are often observed in the ureters, occasionally found in the renal pelvis and bladder. The ureteropelvic junction or upper ureter is the most common location where UFPs are trapped. The median size of ureteral polyps is 4.0 cm and can grow up to 17cm.[2,3] In our case, the size of the ureteral polyp is only 2.0 cm. With this size, diagnosis radiologically is difficult.

The etiology of UFPs is unknown. Many experts believe the disease is congenital, found among children. However, some experts found that chronic irritation and development, such as urinary calculi, were most frequently suspected.[1,2,3] Stones are the most common irritant. Most patients have renal calculi coupled with UFPs. In addition, trauma, hormonal imbalances, and other acquired factors are considered.[1,2,3] UFPs are benign lesions;the recurrence rate of fibroepithelial polyps (FEPs) after resection is very low. Ludwig et al[3] conducted a systematic literature review, describing clinical cases of 134 patients from 1980 to 2014. Only one patient experienced a recurrent after 1-year follow-up owing to incomplete treatment. A review of patients with ureteral polyps between 1950 and 1980 showed that none of the 108 cases had a recurrent after open resection treatment.[6]

No clinical symptoms are observed in the early stage of the FEPs. Although FEPs are rare and reveal a benign feature, patients with ureteral polyps can experience significant discomfort and undergo nephrectomy due to misdiagnosis. Schneider al.[5] found that 17 articles published in the last decades reported 28 cases of ureteral FPs in age groups from 6 weeks to 12 years. Flank or upper abdominal pain was the principal complaint in 86% of the cases. Other symptoms include hydronephrosis, urinary tract infection, persistent proteinuria, pyuria, haematuria, etc. In our case, FEPs caused severe hydronephrosis, persistent bacteriuria, and urine-derived bacteremia, leading to persistent high fever in children, which was gradually controlled after the renal percutaneous nephrostomy. Schneider al.[5] found that FEPs could cause haematuria in 3% of patients. However, Ludwig et al. [3] argued that flank pain and hematuria were the most common symptoms. In adults, hematuria is the most common symptom of malignant tumors. However, most ureteral tumors are malignant, and the proportion of benign tumors is less than 1%. Thus, preoperative diagnosis is important.

Preoperative imaging diagnosis of FEPs is difficult, but ultrasound and MRU are the most common methods for urinary tract investigations in children. Schneider[5] found that the sensitivity of ultrasound, magnetic resonance imaging, and computed tomography (CT) scans for FEPs is low approximately 49% in adults. However, FEPs can be misdiagnosed as upper tract urothelial carcinoma. Intravenous urogram (IVU) and retrograde pyelography are the most ideal preoperative diagnostic methods, in which the rate can be up to 70 percent. However, a smooth filling defect is present in IVU. Enhanced CT does not provide more information than IVU. In addition, a ureteroscope is useful, and FEPs in adults can be diagnosed pathologically. However, the narrow ureter is the main obstacle to the use of ureteroscopy in children.

With limited endoscopic techniques for children and the benign nature of FEPs, pyeloplasty is the preferred treatment option for FEPs, especially for UPJ obstruction. Other treatment options are open laparoscopic or robot-assisted pyeloplasty.[9,10] For patients with hydronephrosis and infection caused by ureteral polyps, active infection control is considered before surgery. since poor infection control can lead to poor anastomotic healing, causing urine leakage. In the present case, right renal percutaneous nephrostomy was performed before surgery; infection was strictly controlled, and anastomosis healed well rapidly. Thus, endoscopic resection of UFPs is a safe and it is an effective option in adults. Under the mature endoscopic technology, endoscopic resection is a minimally invasive surgical method, used as

the gold standard for the treatment of ureteral polyps. Other treatment options are ureteroscopy/holmium laser lithotripsy or percutaneous nephroscopy.[3,7,8,11] Meanwhile, nephrectomy is performed only when renal function is completely lost due to hydronephrosis.

UFPs are a rare primary cause of UPJO. However, UFPs should be considered when the clinical and radiographic findings of the patients are incompatible with the common etiology. Pyeloplasty is an effective treatment method for polyps in children.

Abbreviations

UFPs：Ureteral Fibroepithelial Polyps；UPJO：Ureteropelvic junction obstruction；FEPs：Fibroepithelial Polyps；IVU：Intravenous urogram；CT：Computed tomography；MRU：Magnetic resonance urography; HE: Hematoxylin-eosin staining.

Declarations

Ethics declarations

Ethics approval and consent to participate

Not applicable

Consent for publication

The parents gave their written consent for their child’s personal or clinical details along with any identifying images to be published in this study.

Availability of data and material

Data sharing is not applicable to this article as no datasets were generated or analyzed during the current study.

Competing interests

Authors have no conflict of interests.

Funding

No funding was obtained for this study

Authors' contributions

LXF designed the study;DYT,HDC,ZBZ were responsible for collection of data. All authors have approved the submitted version.

Acknowledgement

The authors gratefully acknowledge American Manuscript Editors for English language editing.

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Figures

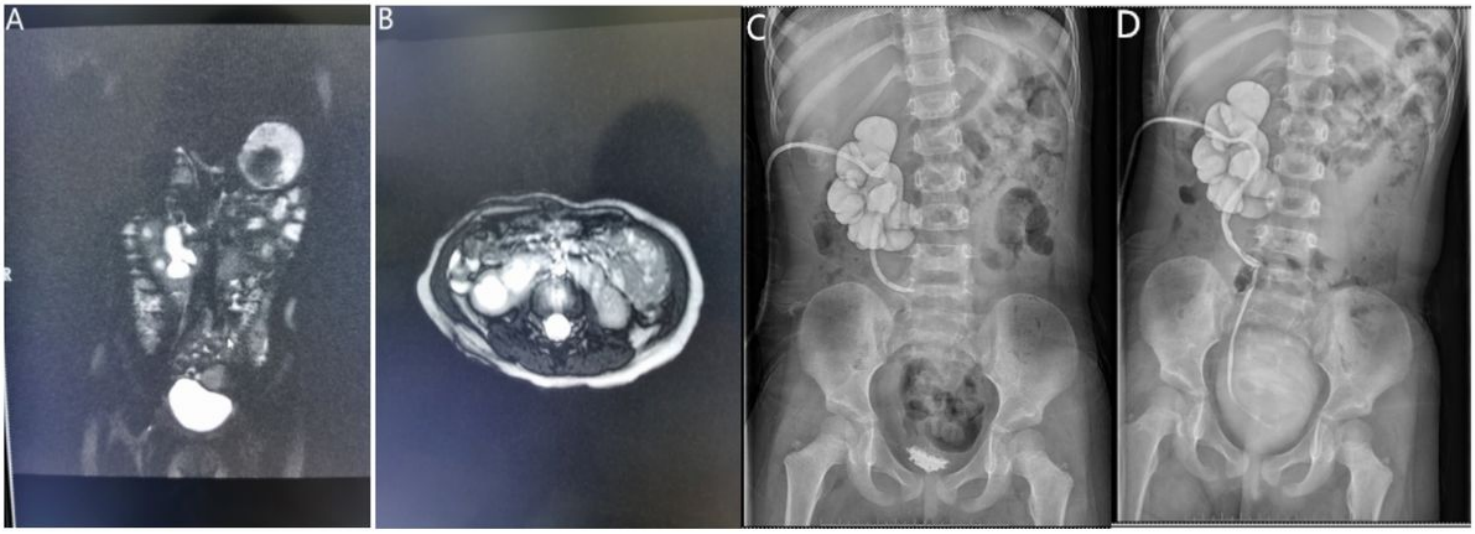

Figure 1

From MRU(A,B), the right kidney is in full shape; the right renal pelvis and calyces are dilated, and the lower kidneys are joined at the midline. From anterograde pyelography (C,D),the nephrostomy tube passes through the ureteropelvic junction; no obvious ureteral stenosis is found.

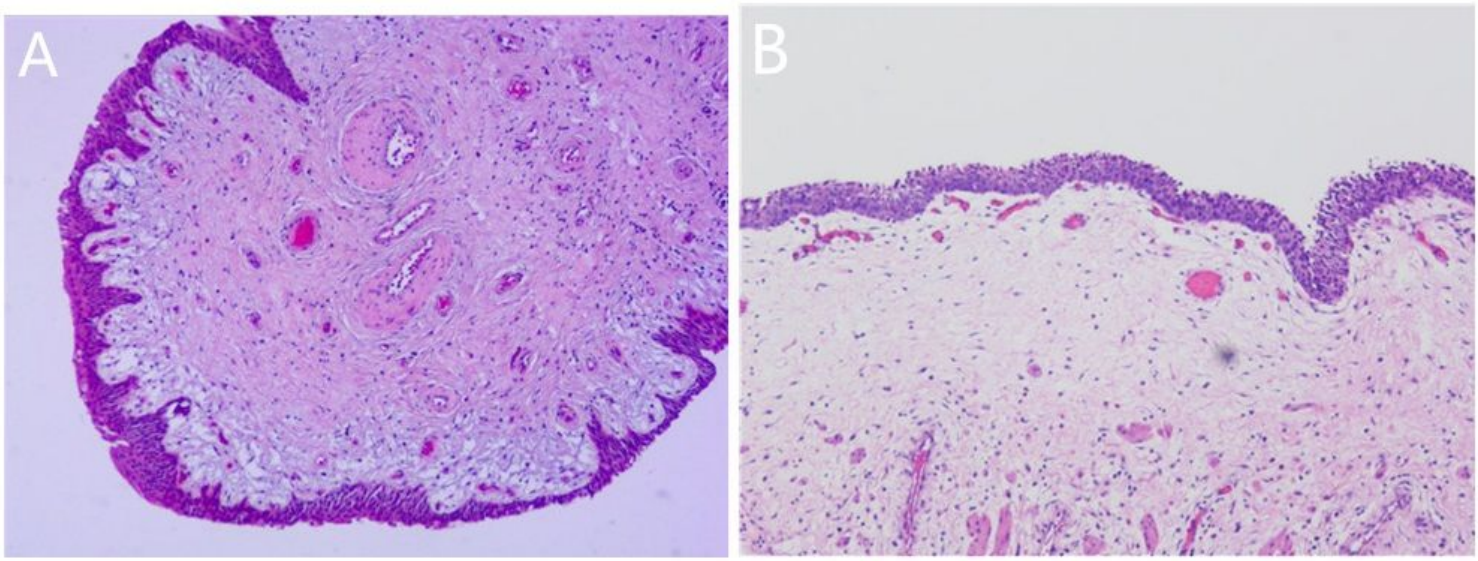

Figure 2

Histological imaging demonstrating fibrous vascular core covered by normal urothelial epithelium; no malignant cells were detected,(A) HE,× 40. (B)HE,× 100.