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CASE REPORT

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Surgical management of high-grade vesicoureteral reflux in an 18-month-old female with a solitary kidney: A case report from a resource-limited setting

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Key Clinical Message

Conservative nonsurgical therapy ensures that the resolution is nearly 80% for vesicoureteral reflux grades I and II and 30%–50% for vesicoureteral reflux grades III and V within 4–5 years of follow-up. Open surgical reimplantation of ureters of grades IV and V is a highly successful procedure, with reported correction rates ranging from 95% to 99% regardless of the severity of vesicoureteral reflux.

Abstract

Patients with vesicoureteral reflux present with a wide range of severity. With an incidence of approximately 1%, vesicoureteral reflux is a relatively common urological abnormality in children. Postnatal diagnosis of vesicoureteral reflux is typically made following a diagnosis of a urinary tract infection and less frequently following family screening. Voiding cystourethrograms remain the gold standard for diagnosing vesicoureteral reflux. To preserve the kidney and prevent the need for potential renal replacement therapy, infants with a single kidney require significantly more assessments and prompt decision-making. Surgical correction is advised for patients with vesicoureteral reflux grades IV and V, while vesicoureteral reflux grades I, II, and III are managed conservatively.

KEYWORDS

High grade vesicoureteral reflux, solitary kidney, ureteric reimplantation, voiding cystourethrogram

1 | INTRODUCTION

Vesicoureteral reflux (VUR) is an anatomical and/or functional disorder with potentially serious consequences, such as renal scarring, which increases with the severity of reflux, hypertension, recurrent pyelonephritis, and renal failure.[1] Many reflux patients do not develop renal scarring and most likely do not require intervention.

Patients with VUR present with a wide range of severity. With an incidence of approximately 1%, VUR is a relatively common urological abnormality in children.[2]

Neonates with prenatal hydronephrosis may have a 15% increased prevalence of VUR,[1] and children with febrile UTIs may have a 30–45% increased prevalence of VUR.[3] To preserve the kidney and prevent the need for potential renal replacement therapy, infants with a single kidney require significantly more assessments and prompt decision-making. Due to anatomical variations, girls experience urinary tract infections (UTIs) more frequently than boys. On the other hand, boys are more likely than girls to have VUR (29% vs. 14%) among all children with UTIs. Furthermore, VUR in boys is more likely to resolve on its own, although they also typically have higher grades of VUR diagnosed at younger ages.[4] Postnatal diagnosis of VUR is typically made following a diagnosis of a UTI and less frequently following family screening.[5]

The goal of the diagnostic work-up should be to assess the child's general health development, renal status, UTI history, presence of VUR, and Lower urinary tract function. A comprehensive physical examination for infants with bilateral renal parenchymal abnormalities or a solitary kidney includes measuring blood pressure, performing urinalysis to determine presence of proteinuria, culture of the urine, and measuring serum creatinine.[6]

The standard imaging tests include kidney, ureter, and bladder (KUB) ultrasound, voiding cystourethrogram (VCUG) and nuclear renal scans. KUB ultrasound and radionuclide cystography (RNC) could be considered complementary modalities.[7] The VCUG remains the gold standard for establishing the presence and degree of VUR.[7]

Refluxing ureterovesical junctions can be anatomically corrected through surgery. The surgical techniques employed are open surgery, endoscopic correction, and robotic-assisted laparoscopic reimplantation[6]. Bilateral extravesical ureteral reimplantation has been associated with postoperative urine retension, often requiring long-term catheterization. However, this is an uncommon occurrence. Patients with grade I, II, and III VUR have a high chance of spontaneous remission and a low-risk of renal scarring; therefore, surgical correction is not advised for these patients. For those with grade IV or V reflux, surgery might be recommended or reserved for patients who are unable to take prophylactic antibiotic therapy or who develop new illnesses while receiving it.[6]

We present the case of a child who presented with low urine output since the age of 4 months. Investigations revealed grade 5 VUR with a dilated megaureter and a solitary kidney. She was surgically corrected after a thorough clinical assessment. At the 3-month follow-up, the patient's initial symptoms had resolved with good surgical outcome.

2 | CASE HISTORY

We report a case of an 18-month-old female with a history of congenital heart disease who presented at our clinic with chief complaints of difficulty in passing urine since the age of 4 months. Her mother reported a normal voiding pattern at birth. Her condition was gradual in onset but progressive in nature and was associated with excessive crying while voiding and low-grade fever. Her mother reported some anuric episodes twice for 2 days prior to admission. On different occasions the patient had been treated for recurrent UTI presenting with persistent fever, with subtle improvements. Recently, the mother reported history of poor feeding and failure to thrive. She also reported normal prenatal history and had a successful spontaneous vaginal delivery with the baby weighing 3.3 kg and Apgar score of 8–10. She was the only child to her mother. The patient had no history of lower limb edema, no facial or abdominal swelling.

On general examination, she was alert, tachycardic, afebrile, and not jaundiced and had no enlarged peripheral lymph nodes. Her vital signs included a BP of 75/40 mmHg and a PR of 135 beats/min with a normal respiratory rate and body temperature. Anthropometric measurements revealed a mid-upper arm circumference (MUAC) of 11.7 cm with features of moderate acute malnutrition.

Abdominal examination revealed asymmetrical abdomen moving with respiration, mildly distended nontender right lumbar region, with normal external genitalia. Systolic heart sounds with increased precordial activity were noted on systemic cardiovascular examination.

Blood workups revealed leukocytosis, moderate anemia of 8.2 g/dL, thrombocytosis of 511,000 $\times$ 10^9/L, biochemistry analysis revealed elevated serum creatinine by 268 µmol/L and BUN of 34 mmol/L with normal serum electrolytes. The levels of inflammatory markers such as CRP and ESR were slightly increased. Urinalysis was positive for leucocytes. Urine culture and sensitivity revealed Proteus mirabilis sensitive to Amikacin.

3 | INVESTIGATIONS AND TREATMENT

KUB ultrasonography showed right-sided renal parenchymal disease with thinned cortex and poor corticomedullary differentiation. A dilated right ureter was visualized with grade 4 hydronephrosis; the urinary bladder was moderately distended and had thickened wall exhibiting double wall sign. Voiding cystourethrography (VCUG) revealed right VUR grade 5 with a severely tortuous ureter and marked dilatation of collecting system. (Figure 1).

Echocardiography revealed perimembranous VSD, mild tricuspid regurgitation and mild right pulmonary artery stenosis with EF of 57%.

Serum creatinine and BUN resolved after 8Fr Foley catheter drainage, and a contrasted KUB CT scan revealed a solitary right kidney with physiological hypertrophy measuring (8.9 cm × 4.53 cm) and markedly dilated pyramids and a pelvicalyceal system. The left renal fossa was empty, and the left kidney was not visualized. The right ureter was severely tortuous and dilated with distal end widening at the UVJ (Figure 2B). After thorough radiological and physical examination, the patient was prepared for surgery to salvage the solitary kidney.

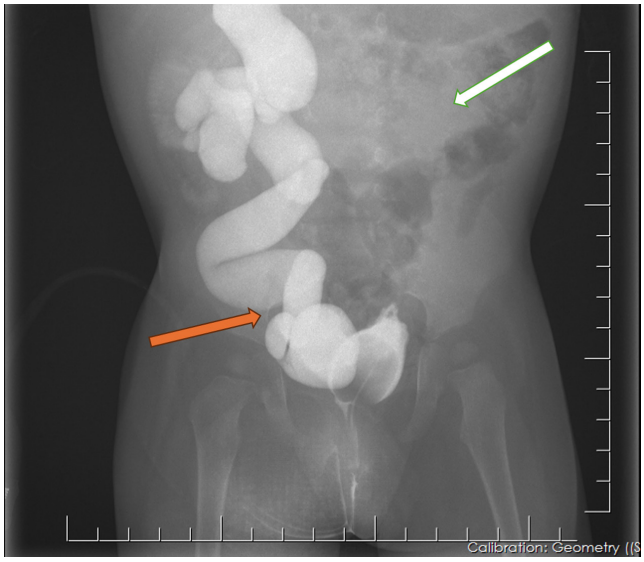

FIGURE 1 VCUG showing the absence of the left kidney (white arrow), right vesicoureteral reflux grade 5 with a severely tortuous of the ureter with marked dilatation of the collecting system (orange arrow).

The abdomen was opened through a sub-umbilical transverse incision via an extraperitoneal approach through the white line of Toldt. The right colon was reflected medially to expose the retroperitoneum. The tortuous ureter was identified on the right anteroposterior part of the bladder (Figure 3). There was an outpouching of the bladder wall at the site of insertion of the right ureter, with abnormal distal ureteric muscle fibers, signaling disruption of the intramural nerve supply, which we believe was the primary cause of the VUR in this patient (Figure 4). The contralateral ureter was not visualized.

The megaureter was mobilized, straightened, and then tapered to facilitate the antireflux mechanism. Approximately 3cm of unhealthy distal ureteric muscle fibers were transected. A DJ stent was inserted, followed by ureteric reimplantation using the Lich-Gregoir extravesical bladder technique (Figure 5), with Vicryl 6–0 sutures.

Abdomen was closed in layers by using Vicryl 2-0 sutures. The patient was placed on a 7-day antibiotic cover, and the DJ stent was removed after 6 weeks. A Foley catheter for urine drainage was left in situ for 21 days.

4 | CONCLUSION AND RESULTS

Surgical repair is typically the best approach for high-grade VUR, as it leads to favorable outcomes and can avoid long-term renal replacement therapy and other serious renal consequences. Our patient, who had a solitary kidney and confirmed high-grade VUR, experienced significant improvement following ureteric reimplantation. Her urine output increased from less than 0.5 to 2.1 mL/kg/h. After 7 days, the control serum creatinine, BUN, and electrolytes were all within the normal range. Three months later, she exhibited a normal voiding pattern with

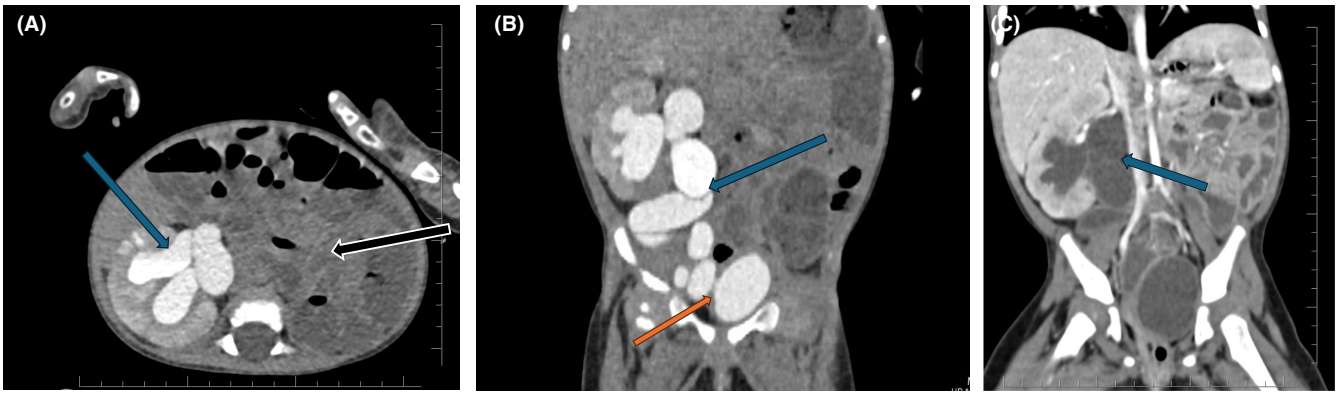

FIGURE 2 (A) Axial view of contrasted KUB CT scan showing a solitary right kidney (on the blue arrow), with physiological hypertrophy measuring 8.9 × 4.53, and marked dilated pyramids and a pelvicalyceal system. Left renal fossa is empty (black arrow) with evidence of left renal agenesis. (B) Coronal view of contrasted KUB CT scan showing severely tortuous right ureter (Blue arrow) and dilated with distal end widening at UVJ (orange arrow). (C) Coronal view of contrasted KUB CT scan showing severely dilated renal calyces and pelvis with grossly dilated ureter (blue arrow).

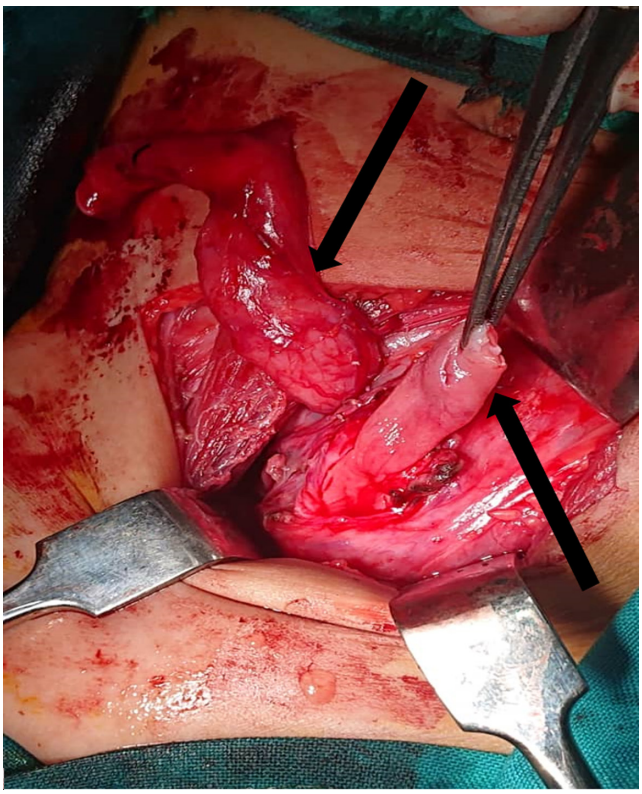

FIGURE 3 Transacted grossly dilated tortuous right ureter inserting superiorly anterior on the bladder wall.

no urological complaints. Although she was scheduled for a follow-up VCUG and KUB CT scan, these tests were not conducted due to financial constraints.

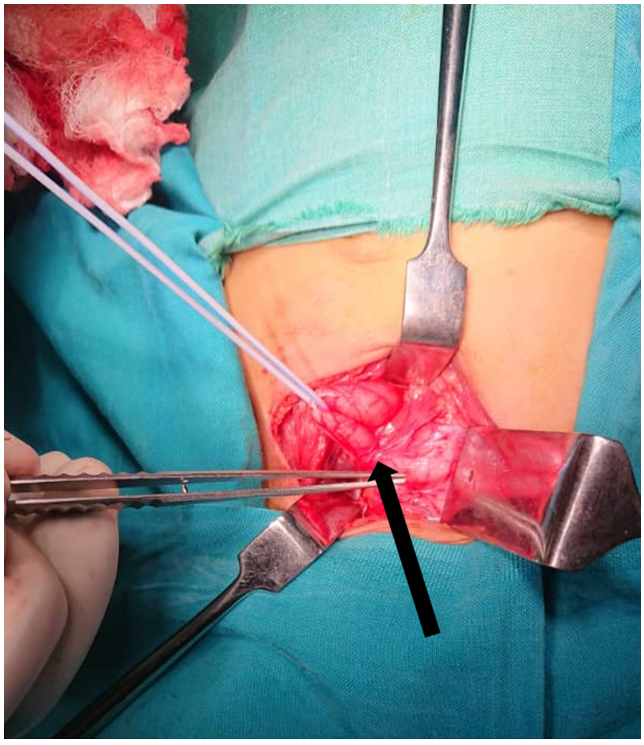

FIGURE 4 Abnormal distal ureteric unhealthy muscle fibers, signaling disruption of the intramural nerve supply.

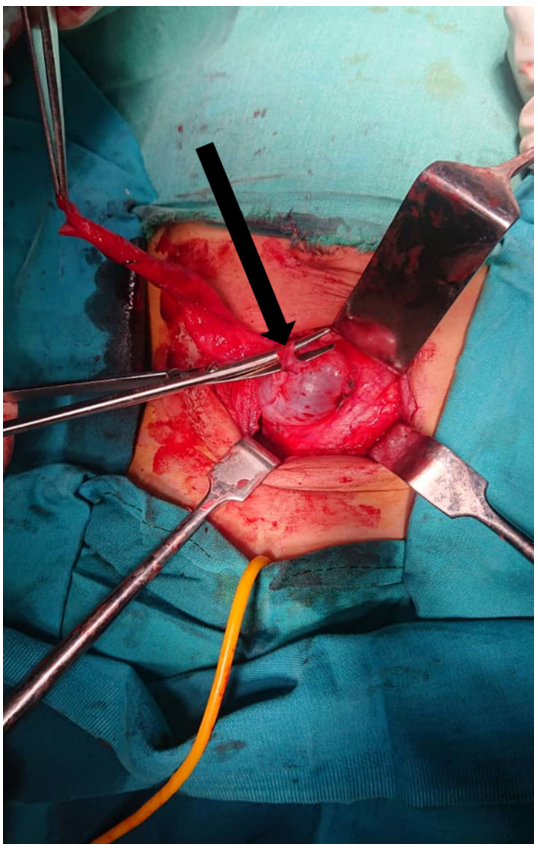

FIGURE 5 Ureteric re-implantation by using the Lich Gregoir extravesical technique.

5 | DISCUSSION

Primary VUR is the most common form of reflux and is caused by incompetent or inadequate closure of the ureterovesical junction (UVJ), which contains a segment of the ureter within the bladder wall (intravesical ureter). Normally, reflux is prevented during bladder contraction by fully compressing the intravesical ureter and sealing it off with the surrounding bladder muscles. On the other hand, secondary VUR is caused by an unusually high bladder voiding pressure, which prevents the UVJ from closing during bladder contraction. It is frequently linked to functional bladder blockage (such as bladder bowel dysfunction [BBD] and neurogenic bladder) or anatomic abnormalities such as posterior urethral valves.[8]

Many children with VUR present with symptoms of recurrent UTIs and a few of them manifest with uremic symptoms such as nausea, vomiting, and hiccups. Comprehensive physical examination, including measurements of height, weight, and blood pressure, is of paramount importance for patients suspected of having VUR

disease. When a child with a first febrile UTI is diagnosed, the following risk factors can be used for the generation of a predictive score for VUR presence: age (>6 months), presence of sepsis, WBC count (>15,000/mm), and abnormal renal USG results.[9]

Laboratory evaluation, such as urinalysis to check for proteinuria, a first-morning urine protein/creatinine ratio should be measured if the urinalysis results are positive for protein, as proteinuria may be a marker of severe chronic kidney disease. Urine culture should be performed on a suitable specimen if pyuria or bacteria are found during a urinalysis. If the child has a single kidney or bilateral renal involvement, serum creatinine should always be measured.[10]

Ultrasound of the kidney, ureter, and bladder is the first standard evaluation tool for children with prenatally diagnosed hydronephrosis. It is non-invasive and provides reliable information regarding kidney structure, size, parenchymal thickness, and collecting system dilatation.[11] Ultrasound should be delayed until the first week after birth because of early oliguria in neonates.[11] The presence of cortical abnormalities on ultrasound (defined as cortical thinning and irregularity, as well as increased echogenicity) warrants the use of VCUG for detecting VUR.[7]

The VCUG remains the gold standard for establishing the presence and degree of VUR because it allows better determination of the grade of VUR (in a single or duplicated kidney) and provides precise anatomic details of the kidney, bladder and urethral configuration.[7] On the other hand, radionuclide studies for the detection of reflux have shown less radiation exposure than the VCUG, but the anatomical details depicted are inferior.[12]

Radionuclide cystogram (RNC) is an alternative modality.[13] However, despite the increased radiation exposure associated with it, VCUG is still a solid option because it offers more anatomic detail. Specifically, RNC does not reliably show a bladder wall appearance, or Grade I reflux.[13] The RNC also does not demonstrate urethral anatomy in boys, which may be important for secondary causes of VUR (e.g., posterior urethral valves). For this reason, in many canters, RNC was not used in the initial study but may be used to monitor for persistent reflux in follow-up studies.

Open surgical reimplantation of ureters is a highly successful procedure, with reported correction rates ranging from 95% to 99% regardless of the severity of VUR.[14,15] In the intravesical approach described by Politano and Leadbetter, the bladder is opened, and the ureters are reimplanted by tunneling a ureteral segment through the detrusor (bladder wall muscle), thereby creating a submucosal tunnel that is long enough to act as a flap valve. Modifications of the basic technique are named after surgeons who developed each of the variants (e.g., Cohen, Glenn-Anderson). In the extravesical approach, reimplantation is performed without opening the bladder and is known as the Lich–Gregoir procedure. If an extravesical procedure (Lich–Gregoir) is planned, cystoscopy should be performed preoperatively to assess the bladder mucosa and the position and configuration of the ureteric orifices.[14] Overall, all surgical techniques offer very high and similar success rates for correcting VUR.[14]

Conservative nonsurgical therapy ensures a resolution of nearly 80% for VUR grades I and II and 30%–50% for VUR grades III and V within 4–5 years of follow-up; however, spontaneous resolution is low for bilateral high-grade reflux.[16] The conservative approach includes watchful waiting, intermittent or continuous antibiotic prophylaxis (CAP), and bladder and bowel rehabilitation in patients with lower urinary tract disease.[10]

There is still disagreement over the best course of action for treating VUR in infants.[6] Most infants with VUR, even those with high-grade reflux, typically outgrow this condition on their own by the time they are 5 years old;[6] however, for patients with solitary kidney presenting with a tortuous megaureter, surgical correction is unavoidable.[17] Regardless of surgical technique, patients may require postoperative bladder drainage via a urinary catheter and in-hospital admission, which usually lasts from one to several days.[15] Our patient had a solitary kidney and was managed by surgical ureteric reimplantation with good surgical outcomes after surgery.

The spontaneous resolution of VUR is dependent on age at presentation, sex, grade, laterality, mode of clinical presentation, and anatomy. Faster resolution of VUR is more likely to occur at less than 1 year of age at presentation, with a lower grade of reflux (grades 1–3) and an asymptomatic presentation of prenatal hydronephrosis or sibling reflux.[18]

Other correction techniques include endoscopic subureteral injection of bulking materials, a less invasive ambulatory procedure, and injection of a periureteral bulking agent via a cystoscope, which changes the angle and perhaps fixation of the intravesical ureter, thereby correcting VUR. The two most commonly used techniques use a copolymer of dextranomer/hyaluronic acid (Dx/HA or DEFLUX) but use different injection sites.[19] The hydrodistension implantation technique involves placing the bulking agent within the ureteral tunnel sub-ureteral transurethral injection, which places the bulking agent outside the ureteral orifice. The success rate for correcting VUR with DEFLUX in one or more procedures ranges from 75% to over 90%.[20]

AUTHOR CONTRIBUTIONS

Charles John Nhungo: Conceptualization; investigation; supervision; validation; visualization; writing – original draft; writing – review and editing. **Kelvin Richard Mwakalukwa:** Conceptualization; investigation. **Erasto Phares Wambura:** Investigation; methodology; supervision; validation; visualization. **Herry Godfrey Kibona:** Writing – review and editing. **Fransia Arda Mushi:** Writing – review and editing. **Nimwindael Stephen Msangi:** Investigation; software; validation. **Isaack Mlatie Maro:** Supervision; visualization; writing – review and editing. **Njiku Marko Kimu:** Writing – review and editing. **Obadia Venance Nyongole:** Supervision; visualization; writing – original draft; writing – review and editing. **Charles A. Mkony:** Supervision; validation; visualization; writing – original draft; writing – review and editing.

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CONFLICT OF INTEREST STATEMENT

The authors declare that they have no competing interests.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICS STATEMENT

This case report study was exempt from ethical approval at our institution, as this paper reports a single case that emerged during normal surgical practice.

CONSENT

Written informed parental consent was obtained from the parents/guardians for publication of this case report and accompanying images. A copy of the written parental consent is available for review by the Editor-in-Chief of this journal upon request. Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

DISCLOSURE

This report has been published in accordance with the CARE criteria.

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