African Urology 2023;3:91-93
https://doi.org/10.36303/AUJ.0080

AFRICAN UROLOGY
ISSN 2710-2750 EISSN 2710-2750

CASE REPORT

Duplicated left ectopic ureter with ureterovaginal fistula in a Nigerian girl

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The purpose of this case report is to relate our experience with the challenges in the management of this extremely rare case. Our report is on an 18-year-old girl who presented with dribbling of urine per vagina since birth. Her voiding pattern was paradoxical. Clinical examination only revealed pooling of urine in the vaginal vault. A suspected diagnosis of a duplicated collecting system with ectopic left ureteral implantation was made following computed tomography (CT) urography. She had upper pole left ureteral tapering and reimplantation. A duplicated collecting system can be asymptomatic. Urinary incontinence in a young patient with associated urinary sepsis should arouse suspicion of this condition. The patient regained continence after the surgery.

Keywords: duplicated ureter, ureterovaginal fistula, ureteral reimplantation, Nigeria

Case report

An 18-year-old Nigerian girl presented at the Surgical Outpatient Department, Urology Division of our centre with a history of continuous dribbling of urine per vagina since birth. Apart from this, there were no other lower urinary tract symptoms. She developed recurrent left flank pain at the age of five, which was often relieved by analgesics. There was a history of repeated urinary tract infection which was treated with oral nitrofurantoin at the source of referral. She had no history of haematuria or loin mass. Her past medical and surgical histories were clinically unremarkable. Clinical examination was only remarkable for pooling of urine in the vaginal vault. Urine culture was sterile. Electrolyte, urea and creatine as well as complete blood counts were essentially normal. An abdominopelvic ultrasound showed the left kidney with a duplicated collecting system, but a normal right kidney. An intravenous urograph showed prompt uptake of contrast bilaterally; however, a duplicated collecting system could not be established (Figure 1). The diagnosis in this patient was confirmed by using contrast-enhanced abdominopelvic computed tomography (CT) which showed a complete duplication of the left collecting system. The lower moiety pelvis and ureter were preserved while the upper moiety pelvis and ureter were markedly dilated and tortuous, extending distally into the pelvis (Figure 2). The patient had a left upper pole ureteral tapering by excisional ureteroplasty and open extravesical stented ureteral reimplantation. Intraoperative findings include a markedly dilated upper pole ureter with a diameter of 4 cm extending into the pelvis laterally to an apparently normal lower pole ureter (Figure 3). This associated megaureter necessitated ureteral tapering by excisional ureteroplasty before reimplantation. There were no postoperative complications. She was discharged satisfactorily on postoperative day five. The patient was already dry on clinic follow-up. Postoperative abdominopelvic ultrasound was essentially normal and urine culture was sterile.

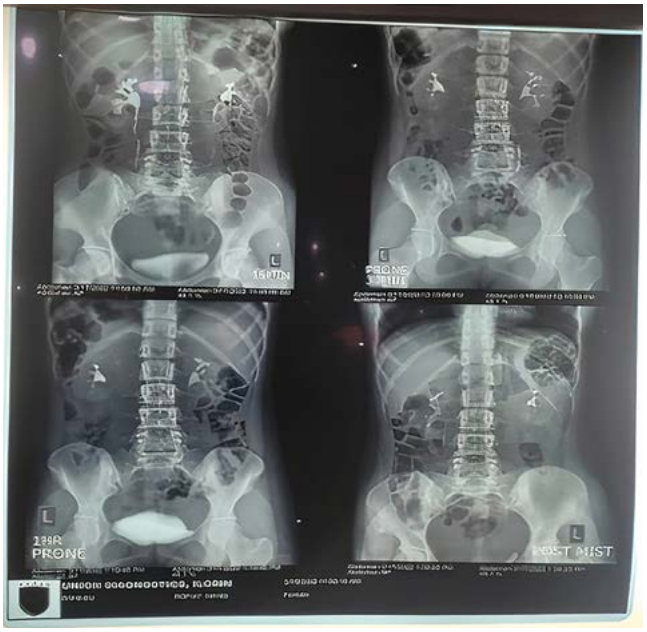

Figure 1: Intravenous urography with prompt uptake of contrast but duplicated ureter could not be established

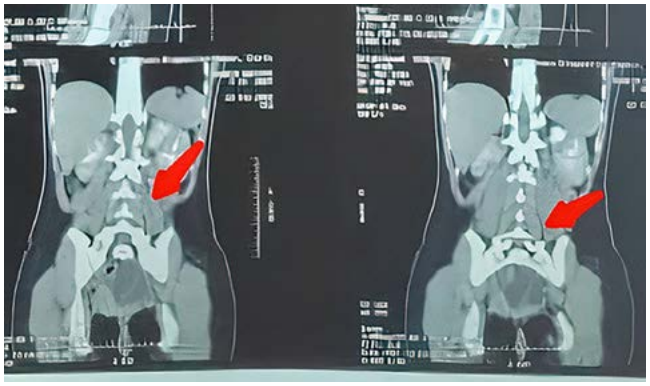

Figure 2: Duplicated left ureter is indicated by the arrow – although it was not distinct, it was later confirmed intraoperatively

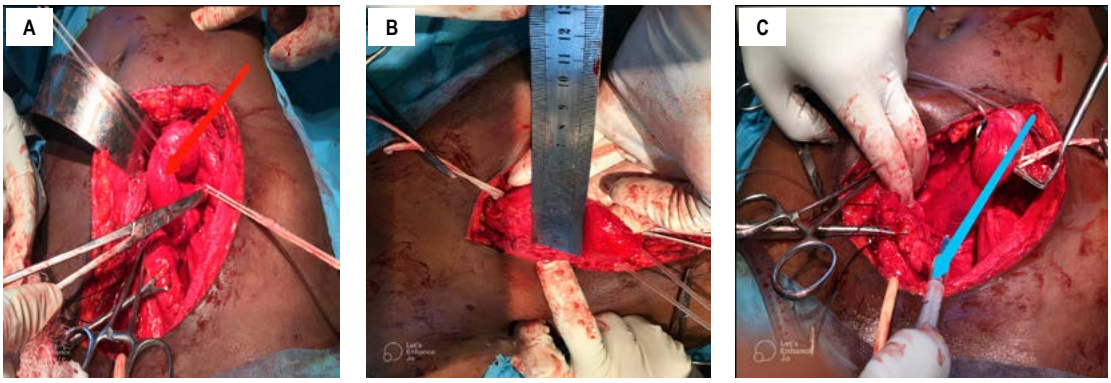

Figure 3: Intraoperative findings showing (A) a markedly dilated and tortuous upper pole ureter; (B) the measurement of the diameter of the tortuous ureter; and (C) the test aspiration of the tortuous ureter with return of urine as indicated by the arrow

Discussion

The ureteric bud arises from the Wolffian duct and gives rise to the development of a normal collecting system through a reciprocal interaction with a metanephric blastema. Disruption in the signalling pathway is thought to be responsible for the abnormal development of the ureteric bud.[1] This includes two ureteric buds arising from the Wolffian duct giving rise to a complete ureteral duplication or one ureteric bud that bifurcates during development, giving rise to an incomplete ureteral duplication.

The incidence of a duplicated collecting system in adults is reported to be 0.7%.[2] It is bilateral in 20% of cases and more common among women.[3] It may also be asymptomatic. However, complete duplication of the upper pole moiety is more prone to complications than duplication of the lower pole moiety. The upper pole moiety may be complicated by hydroureteronephrosis when it is associated with ureterocele.[4,5] This dilation results in an atrophic and non-functional moiety. However, in this reported case hydroureteronephrosis was noticed in the index which may be as a result of ipsilateral vesicoureteral reflux. Upper pole moiety function was largely preserved.

The dilation of the renal pelvis and ureter in this case may be due to partial obstruction because of ectopic vaginal insertion rather than the commonly associated ureterocele. The preservation of some functions of the upper pole moiety, as noted in our case, may not be unconnected to continuous partial ureteral decompression via the ureterovaginal fistula thus limiting renal dysplastic changes.

The observed dilated and tortuous upper pole ureter had an estimated diameter of 4 cm. This may be due to chronic incomplete ureteral obstruction since childhood until the time of presentation at 18 years of age. The lower pole ureter may be associated with vesicoureteral reflux because it may have implanted early compared to the upper pole ureter, leading to a shorter submucosal tunnel which has been linked to urinary reflux.[6] This was, however, not the finding in this case as the lower pole ureter showed no clinical evidence of vesicoureteral reflux.

Urinary incontinence is a significant complication that may be associated with ureteral duplication as documented in the medical literature. This is seen mostly in women due to either infrasphincteric ureteral implantation or ectopic vaginal implantation as it was noted in our case. This is unlikely in men, where most of the ectopic ureteral implantation is above the striated sphincter.

Ectopic ureteral implantation is rare, with an estimated incidence of 0.05–0.025%.[7] The Weigert–Meyer law, which predicts the draining pattern of duplex collecting system, states that in a complete ureteral duplication, the ureter whose orifice is more media and caudal drains the upper renal moiety and the other ureter whose orifice is more lateral and cephalad reaches the lower renal moiety. This law has been observed universally in cases of ureteral duplication and this case was not an exception.

Diagnostic tools for suspected pyeloureteral duplication include ultrasound scan, intravenous urography, voiding cystourethrography, CT and magnetic resonance imaging. Abdominopelvic ultrasound is usually the imaging of choice during the initial assessment of a case of pyeloureteral duplication. It may give an indication toward the diagnosis. However, the assessment of ureteral insertion may be a challenge. Intravenous urography and voiding cystourethrography are complimentary to ultrasonography. In our case, the diagnosis was confirmed by using CT with intravenous contrast. Magnetic resonance urography is considered to be the modality of choice in patients with pyeloureteral duplication due to its superior soft tissue resolutions.[8] This was, however, not done in our patient following conformation with CT scan.

The surgical management of a duplicated collecting system is dependent on related complications at presentation. The aims of management are preservation of renal functions, prevention of urinary tract infection and maintenance of continence. In functioning upper pole renal moiety, such as our case, the procedure that can be done is upper pole ureteric reimplantation. Ureteropyelostomy could also be done in cases where the superior ureter is large and poorly functional with an obstructing ureterocele. In case of a non-functional upper pole renal moiety, an upper pole heminephroureterectomy is done to prevent the risk of infection.

Conclusion

Complete ureteral duplication with congenital ureterovaginal fistula is an uncommon disorder. This case illustrated that a duplicated ureter can be successfully treated even when associated with multiple complications. This procedure improved the quality of life of the patient as she completely regained continence.

Conflict of interest

The authors declare no conflict of interest.

Funding source

No outside funding was required.

Ethical approval

This was waved by ethics and research committee LAUTECH Teaching Hospital Ogbomoso.

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