Association Of Vesico-Vaginal Fistula to The Congenital Ureter Abnormality: About Two Cases in Two African Countries

Yunga Foma Jean de Dieu1*, Venge Mervyn2, Chamutu Maheshe3, Guiro Moussa4, Ogoudjobi O.M5, Tshabu A. Christiane5, Perrin R-X5

1 Obstetrician and Gynecologist - Fistula Surgeon Expert, Higher Institute of Medical Technics (ISTM)/Uvira/Democratic Republic of Congo
2 Obstetrician and Gynecologist senior registrar, Parirenyatwa Group Hospital, Republic of Zimbabwe
3 Resident in Urology department, Faculty of Health Sciences, University of Abomey Calavi
4 General Surgeon and Fistula surgeon Expert, Saint Camille Hospital, Ouagadougou, Burkina Faso
5 Obstetrician and Gynecologist, Faculty of Health Sciences, University of Abomey Calavi, Cotonou, Benin

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*Corresponding author: Dr YUNGA FOMA Jean de Dieu, Obstetrician and Gynecologist - Fistula Surgeon Expert, Higher Institute of Medical Technics (ISTM)/Uvira/Democratic Republic of Congo. Tel: +22996931966; Email: yungafoma@yahoo.fr

Abstract

Background and Purpose

The association of Ureteral duplication to a contralateral ureteral bifidity is an unusual developmental abnormality. A Vesicovaginal fistula associated to the above abnormality has never been reported.

Patients and Observation

We are reporting the first cases of VVF associated to the ureter congenital abnormality. The 1st case is concerning a VVF associated to left ureter duplicity and a right mega ureter identified during a VVF repair procedure whilst the 2nd case is about a VVF associated to a right ureter duplication and a left ureter bifidity discovered during a VVF repair. We discovered the 1st case during an obstetric fistula repair camp at Chinhoyi Provincial Hospital in Zimbabwe. The patient is fully recovered and asymptomatic. The second case is one of the obstetric fistula patients operated at Saint Jean de Dieu hospital of Tanguieta in Benin. Patient still needs another operation to reimplant the bifid ureter into the bladder.

Key words: Fistula; Duplication; Ureter.

Introduction

A significant number of preventable deaths occur during pregnancy and childbirth in developing countries where maternal mortality ranges from 500 to 1000 per 100,000 live births; for every woman who dies, an estimated 16 to 30 more survive with preventable complications. Obstetric fistula can be considered the most tragic complication during childbirth.

WHO estimates that there are about 2 million cases of VVF worldwide and 50,000 to 100,000 new cases each year. [5, 6] The vast majority of cases are reported in sub-Saharan Africa and Southeast Asia where qualified obstetric assistance is random. Obstetric fistulas are thus a major public health problem in poor countries.

Obstetric fistula is an abnormal communication between the urinary tract (the ureter, bladder and urethra) and/or digestive (rectum), on the other hand the genitals (the uterus and vagina) occurring during delivery. Various types of fistula are distinguished: uretero-vaginal fistulas, vesico-uterine fistulas and vesico-vaginal fistulas (VVFs). These take into account uretero-vaginal fistulas. RVF, associated or not with a VVF, is a communication between the rectum and the vagina. The most common are VVF.

Many etiologies are recognized in VVF, but studies in Africa have shown that in the majority of cases, are of obstetric cause. Obstetric Fistula occurs as a result of obstructed labor characterized by prolonged delivery without the possibility of caesarean section.

Uretero-vaginal fistula is characterized by the ectopic drainage of the ureter in the vagina causing a permanent flow of urine through the vagina. Although the congenital origin is rare, iatrogenic etiology remains common during a caesarean section or poorly performed hysterectomy. It results from a ligation or damage of the ureter at its crossing with the uterine artery. The ureter attached to the vaginal wall eventually drains urine into the vagina within a few hours or days of the procedure.

Duplicity and uretero-pyelic bifidity are rare abnormal uropathies whose surgical discovery is the most common diagnostic method in developing countries where high-tech
diagnostic method is not affordable. The rarity of the pathology and the non-systematic prescription of certain analysis (as IVU) for the preoperative assessment of urogenital fistulas justify the paucity of the literature on this subject.

**Objectives**

To describe the rare cases of association VVF - congenital malformation of the ureter, their treatment and prognosis.

**Patients and Observations**

**First Case**

Vesico-vaginal fistula associated to left ureteral duplicity and a ligated right mega-ureter. Case of a patient operated at Chinhoyi Provincial Hospital in Zimbabwe.

Mrs. S.B., married, G6P6Alive5, 43 years old was admitted at Chinhoyi Provincial hospital on 2/21/2018 for leaking of urine for the previous 2 years after a caesarean-section of her 6th pregnancy. Patient reported not to have leaked urine prior to the cesarean section. She had a previous vesico-vaginal fistula repair vaginally done at Gokwe Hospital.

The physical examination showed flow of urine through the vagina with a positive dye test to confirm a recurrent vesico-vaginal fistula. Pelvic ultrasound was not done. The biological balance was normal except for creatinemia at 149.9 mmol/l and urea at 9.21 mmol/l.

Operation was done using the vaginal approach. During the operation 2 VVF holes were noted, the left ureter was draining in the vagina outside the bladder cavity and the absence of the right ureteral orifice.

VVF repair and left ureter catheterization were performed on 2/23/2018. Intravenous urography was prescribed and left ureteral reimplantation was considered as soon as possible.

The postoperative period was normal and the patient was discharged from the hospital on 3/08/2018 without the IVU being done.

She was readmitted on 2/12/2019 for the same complaint. Physical examination revealed a left uretero-vaginal fistula associated with a high vesico-vaginal fistula identified after the methylene blue test. Despite the impossibility of performing IVU within the hospital, it was decided to operate on the patient on 2/18/2019.

During the abdominal procedure, the internal genital organs had normal appearance.

First step: **VVF repair**

At cystotomy, there is no drainage of urine in the bladder and no ureteral meatus but a small vesico-vaginal fistula was discovered. By trans vesical approach, a circular incision was made around the fistula followed by a dissection to mobilize the bladder from the underlying vaginal lining. The bladder wall was closed in one layer and interrupted suture using the 3/0 vicryl suture.

Second step: Discovery of left ureteral duplicity and a ligated right mega-ureter followed by their reimplantation in the bladder (See photos 1 to 5).

On the left side, after opening the posterior peritoneum next, we discovered two similar structures of different sizes making us suspect a congenital ureter’s malformation confirmed after

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**Figure 1:** Isolation of the 3 ureters on the lakes

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their puncture that brings back clear urine. After dissection, the 2 ureters were isolated separately on two lakes, sectioned at their distal ends and catheterized respectively by ureteral catheter size 5 and 6, both draining clear urine.

Right ureter: The exploration of the right side revealed a dilated and tick right ureter. The retro peritoneum is opened followed by a dissection to isolate and mobilize the ureter to its distal end. Isolated on a lake, it was sectioned at its distal end and catheterized with size 16 nasogastric tube.

Trans-vesical tunelizations were done to allow the 3 ureters to be separately re-implanted in the bladder. The ureters are attached to the bladder wall at their entry points and on the bladder mucosa using interrupted sutures of vicryl 3/0. The ureteral catheters were reinserted followed by their transurethral passage. The cystorrhaphy was made in 2 layers with vicryl 2/0 suture. Cleaning the abdominal cavity followed by the closure of the abdominal wall with a suprapubic Foley bladder catheter for 21 days while the ureteral catheters are kept for 10 days.

Figure 2: Catheterization of the right dilated ureter

Figure 3: The duplicated left ureters squatting clean urine
Figure 4: Catheterization of the 3 ureters

Figure 5: Reimplantation of the 3 ureters into the bladder

The postoperative stay was normal and the patient was discharged fully dry and continent. An abdominal and pelvic scan was prescribed but is still to be done.

**Second Case**

*Vesico-vaginal fistula associated to right ureteral duplicity and left ureteral bifidity. Case of a patient operated at St. John of God Hospital in Tanguieta, Benin.*

Mrs. E.M., widow, G11P11Alive3DCD8,50 years old, having been admitted for the first time to the St. John of God Hospital in Tanguieta on 8/07/2019 for permanent involuntary loss of urine after a home delivery of a stillborn birth 9 months prior.

The clinical examination noted a large circumferential vesico-vaginal fistula of 6 cm.

The VVF repair was done on 8/08/2019 but the post-operative evolution was unsuccessful. The patient came back to the hospital on 2/22/2020 for a large recurrent vesico-vaginal fistula. The biological blood test revealed leukopenia at 2.48-10³,
Figure 6: Right ureteral duplicity with distinct visible ureteric catheters in situ

Figure 7: Left ureteral bifidity with left uretero-pyelic dilatation
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GR at 3.91-106, Platelets at 256-103, Hemoglobin at 10.7 g/dl, creatinemia at 7.5 mg/l and a normal ultrasound. During a vaginal VVF repair on 2/27/2020, two ureteral openings were discovered after a furosemide test and were catheterized on the right side. The left ureteral opening was not visualized. Therefore, we suspected a congenital malformation of the urinary tractus. The procedure continues by peri fistular incision followed by a wide dissection to achieve maximum bladder release.

The fistula was closed in one layer by interrupted suture using vicryl 2/0, the vaginal mucosa being sutured with vicryl 0 sutures. The dye test was negative but there was a leak of clear urine noted in the left para-cervical region (cervix) which made us suspect a coexisting left uretero-vaginal fistula.

This required us to request an intravenous urography whose pictures are below confirm the diagnosis of:

- Right ureteral duplicity with distinct visible catheters. Left ureteral bifidity.
- Slight left uretero-pyelic dilatation, versus a grade II vesico-ureteral reflux.

She was readmitted on 7/13/2020 and reoperated on 7/17/2020 for left ureter reimplantation and urethroplastie for stress incontinence. Patient was discharged on 8/22/2020 under perineal physiotherapy for minor post-operative stress incontinence.

Discussion

Several papers have presented different types of ureteral malformation including unilateral ureteral duplication (our second case) but we did not find a case associating ureteral duplication with contralateral ureteral bifidity. Nor were any cases found of vesico-vaginal fistula revealing a urinary malformation.

Smith described in 1946 four types of ureteral triplicity [1]:

- Type I: complete triplicate with three separate ureters and three ureteral holes (35% of cases); Type II: double ureter including a bifid (21% of cases);
- Type III: a single ureter (31% of cases);
- Type IV: two ureters and three ureteral orifices: this is the result of a ureteral duplicity, one of which has an inverted bifidity.

Pyelo-ureteral duplication is a malformative urinary tractus present in 0.6% of the general population [2]

The abnormality is more common to the girls and is sometimes bilateral. Diagnosis is often difficult with paraclinical investigations and surgical discovery is the most common method of diagnosis. Accurate analysis of the abnormality is difficult in the majority of cases on simple intravenous urography because one or more kidney territories may be “mute” or impregnate only late, but it is often surgical exploration that makes it possible to make the diagnosis [3]

A case of triple ureter associated with contralateral duplication has been described by E. Sanchez and Col [4]

The history of urinary incontinence at young age is not found in our two patients to suspect a congenital ectopic ureter abnormality. Nevertheless, CAMPBELL also mentions that almost 50% of women with this abnormality have normal urination, the clinical picture being dominated by the symptoms of urinary tract infection. However, sometimes urine loss is also absent in women and urine loss can begin during childbirth [2].

Conclusion

Although rare, fistula may be associated to a urinary tract malformation requiring special attention during the surgical management steps. Intravenous urography or pelvic scanner may be needed to make the diagnosis.

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