CASE REPORTS

Congenital Vaginal Fistula from a Single System Ectopic Ureter: A case Report

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ABSTRACT

Background: *Vaginal fistulae particularly vesico-vagina fistula, is a common urogynaecologic problem in Sub-Saharan Africa. Majority of these cases are acquired and congenital fistulas are rare. We here report a case of congenital vaginal fistula secondary to an ectopic ureter in 15year old Nigerian female.*

Method: *The case records of a I5year old female who presented to the obstetrics and gynaecological unit of the Federal Medical centre Azare and a Review of literature on the subject was used.*

Result: *A 15-old-girl presented with continuous leakage of urine from the vaginal since birth despite the establishment of normal voiding habit. Examination revealed urine leaking from a pinhole fistula in the vestibule. Intravenous urography showed delayed excretion of the right kidney with a normal single ureter (single system) and normal left kidney. Ureteric implantation into the bladder was performed with good postoperative outcome.*

Conclusion: *In an environment with scarce resources, managing ectopic ureter in the female is quiet challenging. Multidisciplinary approach involving the urologist, paediatric surgeon, radiologist and the gynaecologist will yield the best outcome.*

KEYWORDS: Congenital vaginal fistula; Ectopic Ureter; Ureteric reimplantation.

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INTRODUCTION

Vaginal fistulae particularly vesico-vagina fistula, is a common urogynaecologic problem in Sub-Saharan Africa. Recent data emanating from some francophone countries of the West-African sub-region revealed an incidence of 1.2 per 1000 deliveries for vesico-vaginal fistulae [1]. In Nigeria the incidence of vesico-vaginal is estimated to be 3.5 per 1000 and with an estimated 110,000 new cases occurring each year[2]. Over three-quarter of cases of vesico-vaginal fistula are due to pressure necrosis while the remaining 25% are accounted for by other causes including cultural practices ('Gishiri cuts'), granulomatous infection (genital tuberculosis/schistosimiasis) and malignancy (carcinoma of the cervix)[3]. Congenital causes including Ectopic Ureter are very rare not only in Africa but world over.

Douglas and colleagues[4] defined etcopic ureter as one that opens at the bladder neck or more caudally rather than on the trigone. Embryologically it results from cranial insertion of the ureteral bud on the mesonephric duct, which permits distal migration with the mesonephric duct as it is absorbed into the urogenital sinus. The incidence of Ectopic Ureter is about 1:2000-5000[4] and is associated with duplicate renal systems in over 80% of cases [4-7].

Single system ectopic ureters are not common and usually associated with other congenital anomalies such as anorectal malformations, skeletal anomalities, hypoplasticdysplastic, cryptochidism and cystic ovaries[6,7]. Ectopic ureter is also often associated with hypoplastic/dysplastic kidney[7]. Because clinical problems associated with etopic ureter are more common in girls, only 15% of cases are reported in boys[4]. One third of ectopic ureter in girls open at the level of the bladder neck. Another third opens into the vestibule around the external urethral meatus while remaining third opens into the vaginal, cervix or uterus[4]. In the male, it opens into the posterior urethra in 40% of cases and seminal vesicle in about 60% of cases[4].

The typical presentation of ectopic ureter in girls is that of continuous urinary incontinence despite normal voiding pattern[4-7]. Where the ectopic ureter opens into the bladder neck or urethra it may be complicated with obstruction and reflux and the patient may present with recurrent urinary tract infections[4].

The investigative arsenal available for diagnosing ectopic ureter include ultrasonography, excretory urography, retrograde pyelography, enhanced computed tomography, radioactive scientigraphy (Technetium 99) and vaginoscopy[4-7]. Where excretory urography and renal sonoraphy fail to demonstrate the dysplastic kidney, enhanced CT or technetium 99 scientigrapy has been found to visualize the associated hypoplastic kidney[8-9]. Treatment of ectopic ureter basically involves removal of the dysplastic/hypoplastic kidney (nephrectomy) either via open surgery or laparoscopically[4,5].

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Occasionally where renal function is fairly adequate, reimplantation of the ureter (ureto-cystoneostomy) may be done[6].

We report a case of single system ectopic vaginal ureter in a 15-year-old schoolgirl and managed at the gynaecologic unit of the department of Obstetrics and Gynaecology, Federal Medical Center Azare, Bauchi State, Nigeria.

CASE REPORT

Miss M.B. was a 15-year-old junior secondary school student who presented at the Gynaecologic clinic on the 21st of October 2002 with continuous leakage of urine per vaginum since birth despite the establishment of normal voiding habit. There was no history of genital cutting. She attained menarche three months prior to presentation and bled for 4 days in a cycle of 30 days. Her last menstrual period was on the 13th of October 2002. She was the third in a family of six children. There was no history of similar problem in any of her siblings.

Examination revealed a young healthy looking girl with well-developed secondary sexual characteristics. Her pulmonary, cardiovascular systems and abdomen were normal. She had normal external genitalia and no evidence of genital mutilation. There was urine dribbling from the vaginal orifice. Gentle speculum examination revealed urine leaking from a pinhole orifice located at the junction between the vestibule and the vagina. This orifice accommodated size 6 feeding tube. Bimanual examination revealed normal findings. Rectal examination showed no abnormality. There was no evidence of pelvic masses. An impression of ectopic vaginal ureter was made.

Her packed cell volume was 35% and her hemoglobin electrophoresis pattern was AA. Serum electrolyte and urea were within normal limits. Ultrasonography revealed normal pelvic organs. The left kidney was grossly normal. The right kidney showed poor corticomedullary differentiation and reduction in size. It measured 62mm in its bipolar length. Intravenous urography showed normal left kidney with delayed excretion on the right (up to 12 hours).

At examination under anaesthesia, the ectopic ureter was catherized using size 6 feeding tube. Laparotomy was then performed. The right ureter was mobilized and the catheter was felt inside it. It was traced upwards to the kidney, which was found to be at the normal position but small in size about 6cm in length. When traced downwards it passed beneath the bladder to the vesico-vaginal space to open into the vestibule. The bladder was opened and only the left ureteric orifice was seen. Ureteral re-implantation was then performed with about 3cm of its length in the submucosal tunnel. Postoperative course was uneventful. She was discharged on the 8th postoperative day. She has remained dry and continent.

DISCUSSION

In an environment where vaginal fistulae are common mostly from pressure necrosis and cultural practices and where diagnostic facilities are scarce diagnosing vaginal fistula from ectopic ureter is quiet challenging, It certainly requires high index of suspicion and perhaps experience. The clinical picture in this case was classical continuous incontinence despite successful toilet training[4,7]. As a clinical guide, Zocher[10] and colleagues stated that for a female that has never been continent with constant urine dribbling, the diagnosis of ectopic ureter should be topmost until otherwise proven.

More often than not, ectopic ureter may be diagnosed late (or present late) because the condition may be mistaken to be enuresis[10].

Radiology remains the corner stone for the diagnoses of ectopic ureter. Ultrasonography and pyelography may fail to demonstrate the affected kidney in some cases because of its hypoplastic state. However enhanced CTScan, Magnetic Resonance Imaging (MRI) radioactive (T_{C99}) scientigraphy will visualized the hypoplastic/dysplastic kidney in such cases[8,9]. In this case both sonography and urography demonstrated a single renal system thus showing that the affected kidney had some reasonable degree of function.

The traditional treatment for ectopic ureter is nephroretectomy[4] usually as an open procedure. Recently successful laparoscopic nephrectomy has been reported[5]. However, Waklu and colleagues[6] recommended ureteric re-implantation provided the renal function is judged to be reasonable. In this case the right kidney was preserved, as its functional capacity was fairly adequate.

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