CONGENITAL VESICOUTERINE FISTULA ALONG WITH DISTAL VAGINAL AGENESIS, SOLITARY KIDNEY AND TONGUE TIE: A RARITY

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ABSTRACT: Congenital vesicouterine fistula in association with vaginal agenesis and solitary renal agenesis has been rarely reported. We present a case of 19 year old female suffering from cyclical menouria for last years. Physical examination revealed absence of vagina. IVP revealed left renal agenesis and CT scan revealed left renal agenesis with vesicouterine fistula. Cystoscopy showed vesicouterine fistula located above trigone near midline. Vesicouterine fistula was repaired along with uterine preservation. Sigmoid colon neovagina was created. Patient is doing well and menstruating per vagina till last followup.

KEYWORDS: Congenital, Vesicouterine Fistula, Vaginal Agenesis, Solitary Renal Agenesis, Tongue Tie.

HOW TO CITE THIS ARTICLE: Khalid Mahmood, Amit Kumar, Rana Pratap Singh, Ahsan Ahmad, Takallum Khatoon. "Congenital Vesicouterine Fistula Along with Distal Vaginal Agenesis, Solitary Kidney and Tongue Tie: A Rarity." Journal of Evolution of Medical and Dental Sciences 2015; Vol. 4, Issue 94, November 23; Page: 16007-16009, DOI: 10.14260/jemds/2015/2334.

INTRODUCTION: Vesicouterine fistula is the least common type of urogenital fistula, which is defined as an abnormal communication between the bladder and uterus or the cervix.[1] It has an incidence of 1-4% of all cases of genitourinary fistula.[2] Presently, the main cause of vesicouterine fistula is an iatrogenic injury during cesarean section which accounts for 83-88% of cases.[3,6] Other less frequent causes are inflammatory bowel disease, endometriosis, intrauterine device migration, bladder tuberculosis and congenital causes.[7,8] We report our experience of managing this case surgically.

CASE REPORT: A 19 year old female from Bihar, the Eastern Region of India, presented with complaints of painful cyclical hematuria with clot, ammenorhea, and pain abdomen for the past seven years. Pain abdomen started with cyclical hematuria and subsided with cessation of hematuria. Patient was continent for urine and faeces.

There was no history of urinary tract infection or faecaluria. Patient had undergone operation for tongue tie at the age of four years. Physical examination revealed absence of vagina, although labia minora and majora were adequately developed. Secondary sexual growth such as axillary and pubic hair, breast development were normal. Laboratory examination revealed normal hemogram, liver function tests and kidney function tests. Abdominal Ultrasonography showed normal uterus and ovaries, urinary bladder with solitary left kidney. Intravenous pyelography further documented solitary functioning right kidney.

Financial or Other, Competing Interest: None.
Submission 03-11-2015, Peer Review 04-11-2015,
Acceptance 13-11-2015, Published 23-11-2015.
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DOI:10.14260/jemds/2015/2334.

Contrast enhanced CT scan revealed solitary functioning right kidney along with vaginal agenesis and an abnormal communication between uterus and bladder. Cystoscopy performed during the period of menouria revealed an orifice of size about 8mm in the supratrigonal area in midline. Blood clots were seen coming from the orifice into the bladder. Laparotomy in supine position by low transverse incision revealed well-developed uterus and ovary. Fistulous tract was identified and excised along with closure of uterus and bladder.

A segment of sigmoid colon was isolated on its mesentry and its proximal end was anastomosed to the uterus. The distal end of bowel was brought out via the vaginal introitus and sutured to the skin to create a neovagina. Now she is doing well as per her last followup at one year. She has been menstruating per neovagina.

DISCUSSION: Vesicouterine fistula has been sparsely described in literature. Peak incidence occurs in young woman between 25 and 33 years of age.[3,6,9] The important causes of vesicouterine fistula can be classified as obstetrical, surgical, radiation necrosis, malignancy, inflammatory bowel disease, endometriosis, intrauterine device migration, bladder tuberculosis and congenital lesion. We have reported congenital vesicouterine fistula along with distal vaginal agenesis, which is quite rare.

A case of primary menouria due to congenital vesicouterine fistula with distal vaginal agenesis was reported by Singh V. Sinha R J et al., in 2011.[10] Another case of congenital vesicouterine fistula with vaginal agenesis and left renal agenesis was reported by Erman-Akar M, Ozkan O, et al.[11] Our case was also associated with left renal agenesis along with tongue tie.

Commonest presentation of congenital vesicouterine fistula is cyclical menouria and same was seen in our case. The presence of vaginal agenesis paved the way for the egress of menstrual blood through the bladder. Posteriorly, the

tough Denonvilliers fascias limited its way to rectum. Accurate and early diagnosis of vesicouterine fistula is difficult, since there are many different clinical pictures. In our case, patient was asymptomatic till the age of menarche when she developed pain abdomen and cyclical hematuria.

The mainstay of diagnosis is typical history, physical examination, cystoscopy, and urinary tract imaging.[7] A cystoscopy is necessary to detect fistula, its size, location, proximity to ureteral orifices and inflammatory state of tissue surrounding the fistula. Dye test and hysterosalpingography, which is used in detection of urogenital fistula in noncongenital cause could not be used in this case due to vaginal agenesis. Ultrasound and intravenous pyelography detected solitary right kidney, but fails to detect vesicouterine fistula. Micturating cystourethrogram revealed good capacity bladder without any reflux, but failed to detect vesicouterine fistula.

Additional diagnostic procedure include contrast enhanced CT, MRI, Vaginal ultrasound.[12,8] Our patient underwent contrast enhanced CT scan, which was readily available and affordable to patient. CT scan revealed abnormal communication between uterus and bladder along with left renal agenesis.

Treatment options for vesicouterine fistula include conservative management, medical and surgical therapy.[3,13,14] Surgery is the treatment of choice in most cases.[3,15] In our case of congenital vesicouerine fistula, repair of fistula with vaginoplasty was the aim. Patient had not given consent for hysterectomy, so we did repair of vesicouterine fistula with preservation of uterus. We did sigmoid colon vaginoplasty to create neovagina.

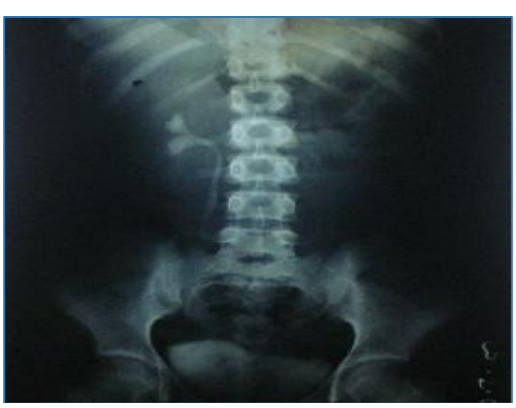

Fig. 1: IVP Showing Solitary Functioning Rt. Kidney

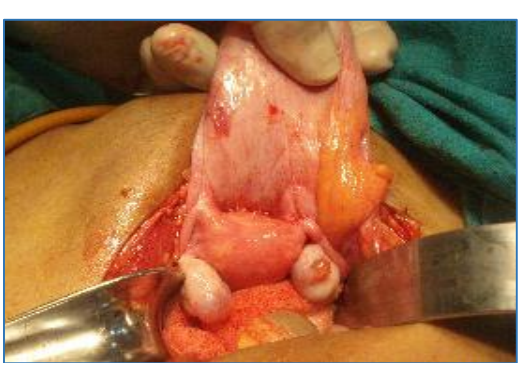

Fig. 2: Intraoperative photograph showing Uterus and Ovary

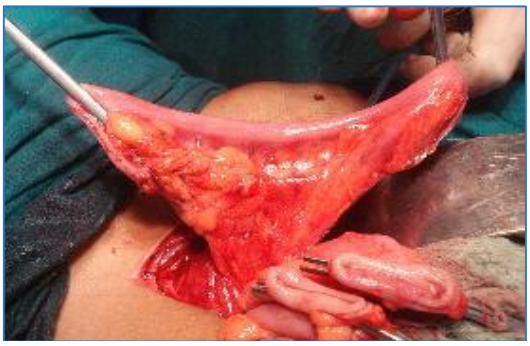

Fig. 3: Sigmoid Colon Isolated to Create Neovagina

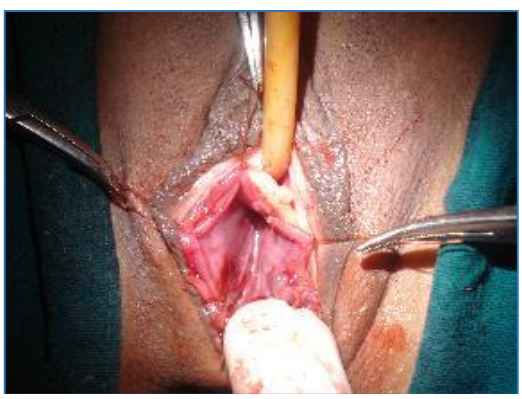

Fig. 4: Capacious Neovagina created by Sigmoid Colon

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