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CASE REPORT

UROGENITAL SINUS ANOMALY: RARE PRESENTATION WITH ACQUIRED RECTOVAGINAL FISTULA IN ADULT FEMALE.

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Abstract:

Urogenital sinus anomaly is a rare congenital anomaly [1]. We report a case of urogenital sinus anomaly in a twenty year old female along with acquired rectovaginal fistula and its systematic management. The patient underwent diversion colostomy, followed by rectovaginal fistula closure, vaginal reconstruction with closure of vesicovaginal fistula. It was followed by colostomy closure one month later. The presentation of urogenital sinus anomaly with acquired rectovaginal fistula is rare and the management is complex.
Key-words: Urogenital sinus anomaly, rectovaginal fistula, adult

INTRODUCTION

Urogenital sinus anomaly is a common communication of vagina and urinary tract anywhere from urethral meatus to bladder, but majority from mid to distal urethra [1]. They exit in the perineum as a single opening [1]. Acquired rectovaginal fistula can occur due to various reasons, they being bowel disorders, iatrogenic causes, obstructed labor, trauma and infections. Our patient presented to gynecology department with complaints of infertility. On evaluation patient was found to have a pure urogenital sinus anomaly and an acquired rectovaginal fistula. This article is sent for publication because of its rarity in presentation, evaluation and multidisciplinary management.

CASE REPORT

A twenty year old female presented with complaints of anxiousness to conceive for one year, cyclical haematuria from puberty [2]. After marriage she had difficulty and pain during coitus. Subsequently she noticed fecal soiling at introitus. Evaluated in gynecology department and found to have a blind ending pouch in the place of vagina, normal urethral meatus below clitoris, fistula between rectum and the blind pouch [Figure 1-a]. Laparoscopy revealed normal uterus, tubes and ovaries. MRI revealed uterus opening into a ballooned vagina, which was found opening into the bladder [Figure-1b]. Fistula between rectum and blind pouch was noted on

clinical examination [Figure 1-c]. Cystoscopy discovered a fistula above bladder neck [Figure 1-d]. So the patient had single urethral opening with confluence of bladder and proximal vagina. A blind pouch posterior to the urethra was present with a fistulous communication to the rectum. The fistula was probably acquired due to intercourse into the pouch or infection.

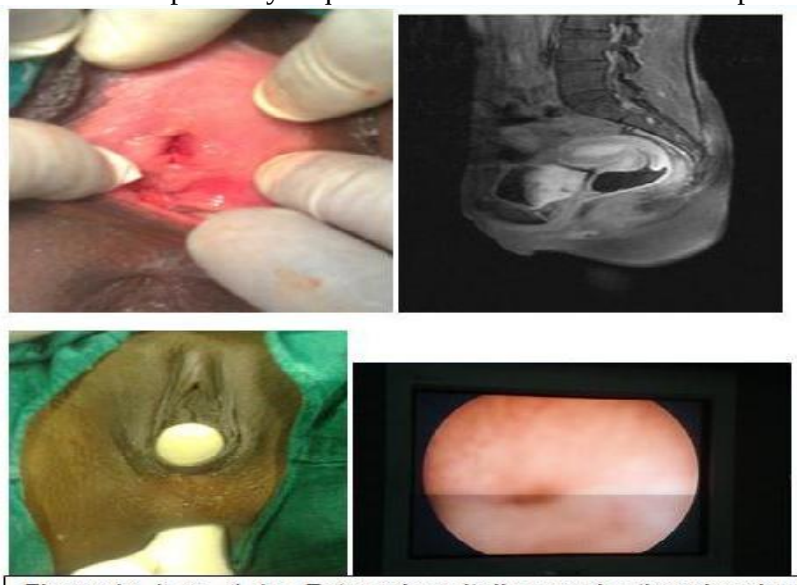

Figure 1a, b, c , d. 1a- External genitalia examination showing blind pouch below urethra 1b- MRI showing ballooned vagina opening into bladder. 1c- External genitalia showing fistula between rectum and blind pouch. 1d- cystoscopy showing vesicovaginal fistula opening in the bladder above bladder neck.

The patient was initially managed by a diversion colostomy. Rectovaginal fistula closure was done. One month later blind pouch was explored. Plane was created between urethra and blind pouch [Figure 2-a]. Proximal vagina opened and was found communicating to the cervix [Figure 2-b]. Vaginal communication to bladder was identified, which was disconnected and closed [Figure 2-c]. Bladder rent was closed. Vagina was reconstructed by pull through vaginoplasty and vaginal wall anastomosed to the external incision made over the introitus. Postoperatively patient was on continuous bladder drainage. Patient menstruated without any discomfort. No urinary leak was present .Colostomy closure was done after four weeks [3].

Figure 2a, b, c. 2-a plane created between urethra and pouch. 2-b blind pouch opened and cervix seen. 2-c vesicovaginal fistula catheterized with ureteric catheter.

DISCUSSION

Embryology [4]: Pure urogenital sinus anomaly is the persistence of the urogenital sinus even after birth. Normally, this common channel undergoes exstrophy and everts to form the vestibule and separates into two separate systems. This procedure fails to occur in urogenital sinus anomaly leading to its persistence.

Rectovaginal fistula can occur due to various reasons, they being bowel disorders, iatrogenic, obstructed labor, trauma, infections or congenital. In our patient it probably was due to sexual trauma or the infection which followed [4].

Cases have been reported which present as variations of cloacal anomaly or incomplete urogenital sinus anomaly. A cloacal anomaly is a condition when the bladder, vagina and rectum communicate and open as a single tube in the perineum. One patient presented with variation of cloacal anomaly with features of communication between bladder and vagina and opening as a single tube and a rectovaginal fistula and anteriorly positioned stenosed anus. Another patient presented as an incomplete urogenital sinus anomaly with urethral type and a blind external pouch indicating incomplete canalization of vaginal plate and partial persistence of urogenital sinus. Our patient presented as a case of pure urogenital sinus anomaly with fistula between rectum and blind external pouch.

In a case of pure urogenital sinus, the various surgical options available are fistula repair followed by vaginal reconstruction. Techniques of vaginal reconstruction include (1) cut back vaginoplasty, (2) flap vaginoplasty (for low vaginal confluence), (3) pull-through vaginoplasty (for high vaginal confluence), (4) complete vaginal replacement (for rudimentary or absent vagina). The other proposed methods are total and partial urogenital mobilization, where the entire sinus is dissected circumferentially and mobilized towards the perineum

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