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Case Report

Laparoscopic uterovaginal anastomosis in bicornuate uterus with cervical and vaginal aplasia

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ABSTRACT

The presence of Cervical and Vaginal Aplasia with bicornuate uterus is a very rare mullerian anomaly. Its true incidence is still unknown. The presence of functioning bicornuate uterus poses a great challenge for a gynecologist because a successful repair could restore normal menses and may preserve a patient's fertility. Hence, we report a case of 14-year-old unmarried female, known case of Bicornuate uterus with cervical and vaginal aplasia with history of a rudimentary horn excision. On clinical and radiological evaluation, she was diagnosed with complete cervical and vaginal aplasia with haematometra in right cornua of uterus. She underwent vaginoplasty along with laparoscopic uterovaginal anastomosis in an innovative way. On follow-up, hysteroscopy showed a patent anastomosis, vagina was completely healed, and she was menstruating normally.

Keywords: Cervical aplasia mullerian anomaly, Uterovaginal anastomosis, Vaginal aplasia

INTRODUCTION

Cervical and Vaginal Aplasia in the presence of uterus is a very rare condition. Its true incidence is still unknown.[1] The prevalence of vaginal agenesis is 1 in 4000-5000 live female births, whereas that of cervical agenesis is 1 in 80,000-10,0000 live births.[2,3] These patients are phenotypically females with normal female genotypes and normal endocrine status.

Most of these patients have small rudimentary uterus without any endometrial cavity, but very rarely unilateral uterine horn may be present with functioning endometrial cavity, with opposite side rudimentary horn.

The successful surgical approach for this condition is neovagina creation, followed by transabdominal approach to create a communication between uterus and vagina by the application of stents. Here we report a rare case managed with an innovative surgery.

CASE REPORT

A 14-year-old unmarried female admitted as a known case of Bicornuate uterus with cervical and vaginal agenesis with history of Left rudimentary horn excision 2 months back. On admission, her vitals were stable with pulse 80 beats/min and blood pressure 120/80 mmHg on the right arm. Her skeletal, cardiovascular, and respiratory system examinations were normal. Her secondary sexual characteristics were appropriate for age with breast tanner stage IV and pubic hairs tanner stage IV. On abdominal examination, no mass was palpable but deep tenderness was present. Pelvic examination revealed vaginal dimple without any vaginal orifice. On rectal examination, no mass was palpable. Her complete blood counts, serum sodium, serum potassium, and random blood sugar were within normal limits. Her serum creatinine was 0.6 mg/dl. Her abdomen and pelvis sonography revealed endometrial collection suggestive of hematometra with Bilateral adnexa normal. Her magnetic resonance imaging (MRI) abdomen-pelvis also revealed

the presence of hydrometra in blind-ended right cornu with absent left cornu, keeping in with history of previous resection and cervical and vaginal aplasia. Both her ovaries were normal and were at the level of pelvic brim. Both of her fallopian tubes were normal. Both kidneys were normal. Her karyotyping was normal with 46 (XX) chromosomes.

Patient's guardians were duly counseled regarding the surgical methods, possible complications, and future fertility aspects, following which a decision of neovaginal creation and uterovaginal anastomosis was taken. On laparoscopy, (Figure 1) there was right side non-communicating functional horn of uterus with right-sided fallopian tube and excised left-side rudimentary horn with normal left fallopian tube. Bilateral ovaries were normal.

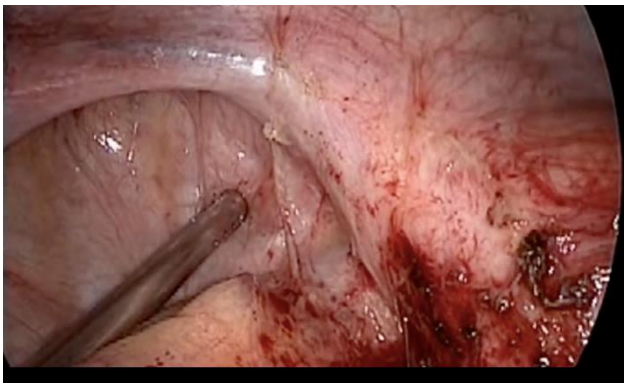

Figure 1: Laparoscopic view non-communicating uterine horn with blind end.

A stab incision was given on the uterus (Figure 2) and hemorrhagic collection was drained. An instrument was passed through the fundus to identify the lowermost pole of uterine cavity.

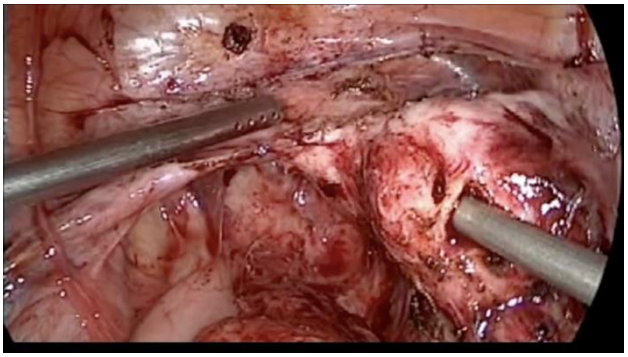

Figure 2: Laparoscopic view stab incision on uterine horn.

A Foley's catheter was placed as a stent between vagina and uterine cavity. Uterovaginal anastomosis was made over the Foley cathetar and vagina reconstructed with Labia minora flaps. Uterine incision was closed with suture. Foley's catheter bulb inflated with 3ml normal Saline. The placement of Foleys catheter in this way served as a communication channel between uterus and vagina. She was started on oral contraceptive pills for 2 months. A follow up hysteroscopy was performed at 8 weeks after surgery. It showed a patent anastomosis with communicating normal uterine cavity and vagina was completely re-epithelialized. (Figure 3). As a result, the patient is having regular spontaneous menstruation with no more complaints of lower abdominal pain.

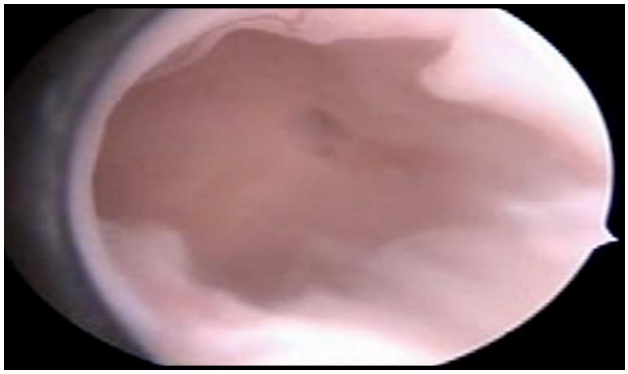

Figure 3: Hysteroscopic view of uterine cavity, endocervical canal with right tubal ostia.

DISCUSSION

Complete aplasia of vagina and cervix with a functioning uterus is an extremely rare malformation.[4]. Very few cases of such abnormality have been reported along with their surgical procedures. Normal vaginal development is by fusion of mullerian duct and the urogenital sinus. Upper 2/3 of the vagina is formed by mullerian tubercle and lower 1/3 by urogenital sinus. Mullerian agenesis can be partial or complete. Partial mullerian agenesis is rare, characterized by normal uterus and small vaginal pouch distal to the cervix, whereas complete mullerian agenesis (MRKH syndrome) is the most common variant encountered. Vaginal agenesis is most commonly associated with MRKH syndrome.[5,6] These patients may have a rudimentary horn or total absence of uterus. Their ovaries are normal, and the secondary sexual characteristics develop normally.

Diagnosis is usually done by typical history of primary amenorrhea with cyclic lower abdominal pain. Clinical examination easily eliminates hymen imperforation. Transabdominal or trans-perineal ultrasonography may specify the level of obstacle and agenesis but are not very reliable for the diagnosis. MRI seems to be the most reliable examination for the diagnosis of uterovaginal malformation and other associated malformations.

Previously, the recommended treatment of cervico-vaginal agenesis was hysterectomy because of high failure rate of canalization procedures and risk of serious ascending infection. But this approach has been replaced by conservative approach due to recent advances in minimal surgery.[7] This surgery should be done in

adolescence period as to avoid longstanding complication of hematometra and hematosalphingnx, which can lead to severe endometriosis, further hampering fertility potential.[8] However, even after such surgery, patients on long term are at risk of stenosis, necessitating further surgery. Repeated episodes of hematometra, endometriosis, and recurrent obstruction may require hysterectomy as a last option. But Laparoscopic surgery of uterovaginal anastomosis should always be a first line of treatment.

CONCLUSION

Cervicovaginal agenesis in the presence of uterus is a rare condition. Preservation of the uterus should always be attempted in such patients. Uterovaginal anastomosis by a minimally invasive approach should be offered as a first-line management for such cases over other treatment modalities with high complications. There is always a possibility of pregnancy either spontaneously or through assisted reproductive technology.

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