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Case Report

Laparoscopic uterovaginal anastomosis for cervicovaginal agenesis

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ABSTRACT

Cervico-vaginal agenesis is an extremely uncommon mullerian abnormality. Therefore, we describe a case of a female patient, age 21, known case of cervico-vaginal agenesis presented with cyclic abdominal pain and vomiting. She underwent three surgeries for vaginal correction to drain haemometra. She underwent Laparoscopic uterovaginal anastomosis. After surgery, the patient experienced regular menstrual cycles without dysmenorrhea. On follow-up, hysteroscopy showed a patent anastomosis, vagina was completely healed and she continued to have regular menstrual cycles.

Keywords: Cervico-vaginal agenesis, Laparoscopy, Menstrual cycle, Uterovaginal anastomosis

INTRODUCTION

In adults, the Mullerian duct forms the uterus, cervix, fallopian tube, and upper part of the vagina. Anomaly in the Mullerian duct is found in about 4.3% of the general population.[1] Cervical agenesis, a rare congenital anomaly, has an incidence of 1 in 80,000 to 1 in 100,000.[2] It is also sometimes associated with partial or complete vaginal agenesis, which is even rarer, reported in 39% of cases of cervical agenesis.[3] Primary amenorrhea and/or cyclical abdominal discomfort are typical symptoms of cervicovaginal agenesis that first appear around menarche. In these situations, the endometrium continues to function normally, but there is no way for the menstrual blood to leave the vagina, which causes hematocolpos and then hematometra to form. If left untreated, the illness may worsen and develop hematosalpinx and endometriosis. [4] Due of its excellent accuracy and thorough delineation of uterovaginal anatomy, MRI is the preferred test for evaluating Mullerian duct anomalies.[5,6] Extensive endometriosis, which in severe cases damages reproductive capacity irreparably and demands salpingectomy and hysterectomy, may develop as a result of delayed diagnosis and treatment.[7] Due to the difficulty of the procedure and the rarity of the anomaly, treatment choices are debatable and include conservative surgery, uterovaginal anastomosis, reconstruction of the endocervical canal, and recanalization of the endocervical canal with a catheter.[8] Due to the high failure rate of canalization treatments, risk of a serious and occasionally fatal ascending infection, and ongoing low fertility, hysterectomy has traditionally been considered in these situations.[9,10] Modern assisted-reproduction facilities and improvements in laparoscopic methods make conservative management both possible and advised. These methods include creation of a neovagina and reconstruction of the cervix around various stents. Herein is presented a case of Cervicovaginal agenesis managed by laparoscopic-assisted uterovaginal anastomosis.

CASE REPORT

A 21-year-old girl presented at the gynecology emergency department with vomiting and abdominal pain worsened

after menstruation and did not improve with analgesics. Five years ago, she had history of cyclical abdominal pain and was diagnosed with cervicovaginal agenesis. She underwent three surgeries for vaginal correction to drain haemometra.

Table 1: Important findings of Cervical-vaginal agenesis case.

Parameters	Observations
Age	21 years
Past history	3 surgeries for vaginal correction within last 5 years
Physical examination	Normal secondary sexual profile, soft abdomen without any organomegaly
MRI finding	Cervical-vaginal agenesis
Surgery	Adhesiolysis, Cervicovaginal anastomosis from the anterior and posterior ends of the cervix at the 3 o'clock, 6 o'clock, 9 o'clock, and 12 o'clock positions
Post operative findings	Follow-up appointments at 1, 3, and 6 months after surgery. The patient's menstrual cycle was regular, and 10 months later, she had no symptoms.

The first two surgeries were performed 4 years ago, and the most recent surgery was done six months ago. Following the vaginal incision and catheter placement for hematometra drainage, the patient developed septicemia with multiorgan involvement 15 days post-surgery. The severity of the condition required a 15-day ICU admission. After six months, the patient was referred to Jivanyog Women's Hospital and Advanced Laparoscopy Centre in Visnagar for further management.

The patient, who had been married for three months, menarche at the age of 12 years. She had a history of a 30-day menstrual cycle lasting for 3 days. She doesn't have any significant personal, past, or family medical history, and there was no diethylstilbesterol exposure during pregnancy. A general physical examination revealed normal secondary sexual profile (breast - Tanner stage 4, pubic hair - Tanner stage 4) as well as average build. Breast and thyroid examinations were normal. An examination of the abdomen revealed a soft abdomen without any organomegaly, although deep palpation revealed discomfort in the lower abdomen. With the catheter in place, pelvic magnetic resonance imaging (MRI) revealed a slightly enlarged uterus measuring 8.9 cm by 6 cm by 4.2 cm. Only a small portion of the cervix was visible, and the upper two thirds of the vagina were absent. The size, position, and appearance of both ovaries were all normal. Cervical-vaginal agenesis was tentatively diagnosed based on these imaging results.

After receiving informed consent, the patient underwent laparoscopic surgery under general anaesthesia. Multiple intestinal adhesions were found during the procedure, possibly as a result of prior procedures or an infection. The adhesions were separated using adhesiolysis.

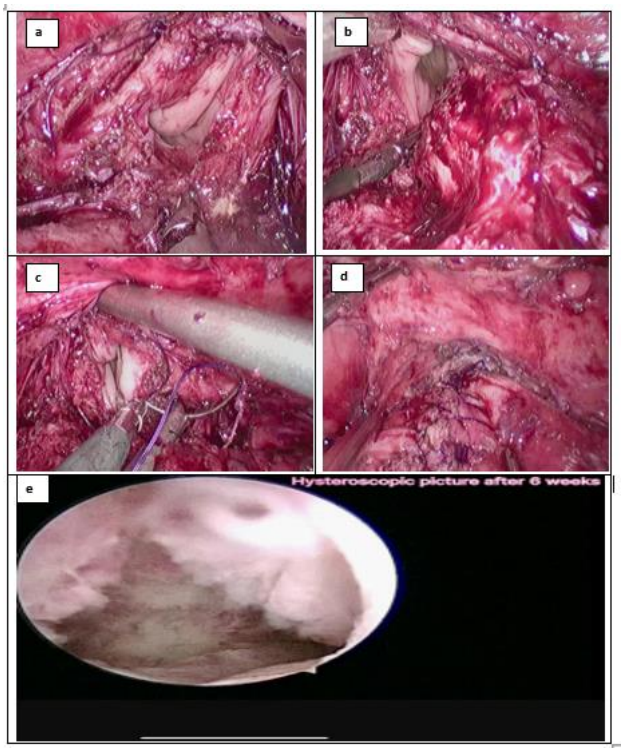

Figure 1: Laparoscopic uterovaginal anastomosis; a) non communicating vagina pouch, b) non connection between vagina and endometrial cavity, c) utero vaginal anastomosis, d) final result after anastomosis, e) Hysteroscopic picture after 6 weeks.

The bladder was divided and adequately pulled down. To expose the posterior portion of the vagina for anastomosis, the rectovaginal space was dissected. To identify the cervical end, a cut was made in the vagina. From the cervical end to the fundus and up to the endometrial cavity, a cervical incision was made. To keep the canal open, a silicone catheter was inserted into the vagina up to the uterus' fundus. Vicryl no. 1 sutures were used to conduct cervicovaginal anastomosis from the anterior and posterior ends of the cervix at the 3 o'clock, 6 o'clock, 9 o'clock, and 12 o'clock positions. Using Stratafix sutures, the vertical incision was closed while maintaining the catheter's position. In the bladder, a drain and an SR Foley's catheter were inserted.

The patient was kept nil orally for 24 hrs and then given a liquid diet for 1 day. They were allowed for solid food on the third day following the procedure. The patient was told to utilise a soft mould for the following 15 days before switching to a hard mould. Intraoperative and postoperative complications were not observed. She has now undergone three follow-up appointments at 1, 3, and 6 months after surgery. After one month, hysteroscopy revealed bilateral ostia and a normal, patent uterine cavity. After surgery, the patient's menstrual cycle was regular, and 10 months later, she had no symptoms.

DISCUSSION

The female genital tract can develop cervicovaginal agenesis with a functioning uterus, which is a rare disease. Only few cases reported in the literature.[11-15] Mullerian duct and urogenital sinus union results in normal vaginal development. The mullerian tubercle makes up the upper two thirds of the vagina and the urogenital sinus the lower third. Partial or full Mullerian agenesis are both possible. While complete mullerian agenesis (MRKH syndrome) is the most frequent type seen, partial mullerian agenesis is unusual and is characterised by a normal uterus and a small vaginal pouch distal to the cervix. MRKH syndrome is most frequently linked to vaginal agenesis.[16] These patients may not have uteri at all or only have a rudimentary horn. Their secondary sexual characteristics mature regularly, and their ovaries are healthy.

Primary amenorrhea, cyclical abdominal pain, and hematometra are the most typical symptoms. In certain situations, retrograde menstruation may cause the development of endometriosis.[17] It is important to differentiate this condition from imperforate hymen, vaginal agenesis, or transverse vaginal septum before initiating definitive management. Vaginal atresia or an imperforate hymen are eliminated by a clinical examination, while cervical atresia and a high vaginal septum may occasionally be difficult to distinguish, necessitating MRI analysis.[18] An related renal tract anomaly can be found with MRI, which is the gold standard for diagnosis.[19]

Cervicovaginal agenesis instances were initially treated with hysterectomy or cervical canalization. Sepsis, endometriosis, and the necessity for additional procedures as a result of restenosis are risks associated with reconstructive surgery in 40-60% of instances.[19,20] Endometriosis also reduces the likelihood of spontaneous conception, frequently necessitating a hysterectomy in the end. But with improvements in surgical techniques and the development of laparoscopy, a minimally invasive method might be the best course of action.[11,21]

Early uterovaginal anastomosis procedures involving cervical drilling or catheterization have substantial recurrence rates and difficulties. In cases of agenesis of the cervix, uterovaginal anastomosis via laparotomy or laparoscopy has been successful, according to numerous publications.[1,7,9,20,22,23] According to Deffarges et al analysis of 18 women who had undergone conservative surgical treatment for uterovaginal anastomosis, all of the women experienced reduced dysmenorrhea, normal menstruation, and no postoperative endocervical canal occlusion.[8] Only one patient out of 18 required additional canalization surgeries due to recurrent uterovaginal canal stenosis, resulting to salpingo-oophorectomy for pyosalpinx. Ten of the 18 patients (56%) experienced spontaneous pregnancies and delivered at term (36-38 weeks) via elective caesarean section. With this method, the likelihood of bladder and rectum injury is decreased, and the uterovaginal canal is reliably canalised.

In 2008, Fedele et al report that cervical atresia was successfully treated by laparoscopy in 12 individuals with vaginal aplasia.[24] Regular menstrual cycles were experienced by all women. The average vaginal length at the 6-month follow-up was 6 cm, and all patients had neovaginal epithelium that was at least 80% iodine positive. During hysteroscopic examination, they verified that all of the patients had mucus at the location of the uterovaginal anastomosis. This recently formed mucus may help transport sperm and play a part in preventing genital infections. Additionally, these women with preserved ovaries and uterus could be able to conceive with the aid of assisted reproductive procedures.[25]

A 10-mm dilator is used to catheterize the uterine chamber during the laparoscopic Deffarges operation, and the atretic tissue is removed to allow for cervical conization.[8] Darwish conducted laparoscopic catheterization, perforated the uterus of six individuals with a sharp-ended probe to insert a Foley catheter inside the cavity, causing excellent menstruation.[26] A relatively innovative method of treating cervical agenesis involves laparoscopic complete excision of atretic cervical tissue and uterovaginal anastomosis. Our method has the benefit of restricting atretic cervical tissue while maintaining healthy uterine tissue.

Without endangering the endometrium, the catheter is introduced into the uterine cavity. Because the atretic tissue is completely eliminated, the freshly created canal is a genuine new endocervical canal, as opposed to laparoscopic total removal of atretic cervical tissue and uterovaginal anastomosis is a relatively new technique for treating cervical agenesis. Our technique has the advantage of setting limits on atretic cervical tissue and preserving normal uterine tissue. The catheter is inserted into the uterine cavity without damaging the endometrium. Due to the complete removal of the atretic tissue, the newly created canal is a real new endocervical canal, as opposed to the Darwish approach where the new endocervical canal is simply a new epithelium covering the Foley catheter.

The size of the newly formed ostium, the length of the newly formed endocervical canal, the presence of vaginal mucosa next to the neo-ostium's end, and the length of stenting all have a role in the success of uterovaginal anastomosis.[27] To prevent vascular or nerve damage, laparoscopic uterovaginal anastomosis requires detailed anatomic understanding of the retroperitoneum. There have been reports that the stenting lasts anywhere from three weeks and three months. In our case, patient is currently free of the symptoms of her menstrual cycle. After 6 weeks, her stent (a Foleys catheter) spontaneously discharged itself, and 9 months after the treatment, she is symptom-free. The possibility of a spontaneous pregnancy as a result of canalization seems very low. Associated endometriosis, pelvic adhesions, and a lack of normal

endocervical canal glandular function may all contribute to this low fertility rate. There have been very few reports of spontaneous pregnancies following canalization of an entirely or partially atretic cervix.[8] For cases of infertility, assisted reproductive technologies may be considered. It is advised to deliver through caesarean section in all pregnancies.[8]

CONCLUSION

When a young girl has primary amenorrhea and cyclical abdominal pain, it's important to consider cervicovaginal agenesis. Early diagnosis and treatment are crucial for a successful outcome. Accurate imaging of the pelvic anatomy is required to avoid complications. Uterovaginal anastomosis is preferred over canalization to reduce the likelihood of secondary cervical stenosis. Early surgery is recommended to prevent complications like endometriosis. Regular menses have a positive psychological impact on these young women.

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