Laparoscopically assisted uterovaginal canalization and vaginoplasty for patients with congenital cervical and vaginal atresia: a step-by-step guide and long-term outcomes

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Research

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Abstract

Objective: To study the long-term outcomes of laparoscopically assisted uterovaginal canalization and vaginoplasty in patients with congenital cervical and vaginal atresia and to introduce the surgery step-by-step.

Methods: A prospective observational study was conducted including 10 patients diagnosed with congenital cervical and vaginal atresia underwent laparoscopically assisted cervicovaginal canalization between January 2016 and June 2020 in a tertiary teaching hospital. Clinical characteristics and perioperative data were recorded. Patients were followed up in outpatient clinic at 3, 6 and 12 months postoperatively, and once a year thereafter. Menstrual cycle and degree of dysmenorrhea were recorded. Gynecological examination was performed to measure vaginal length and to examine whether there was restenosis.

Results: All procedures went smoothly, with no case requiring conversion to laparotomy or no intraoperative complications occurred. Postoperative febrile morbidity occurred in one patient (1/10, 10%). The median (quartile) follow-up time was 26.0 (21.3, 48.3) months. All patients resumed menstruation, including nine patients (9/10, 90%) with regular monthly menstruation. Eight patients (8/10, 80%) experienced mild-to-moderate dysmenorrhea; the remaining 2 patients (2/10, 20%) had no dysmenorrhea. Cervical restenosis occurred in one patient (1/10, 10%) 12 months postoperatively, and cervical dilation was performed. So far, eight months after the second surgery, no restenosis has been found. The mean postoperative vaginal length was 7.9 ± 1.3 cm by the time of last follow up. Only one patient prepared for pregnancy for two years, but she had not conceived yet.

Conclusion: Laparoscopically assisted uterovaginal canalization and vaginoplasty is an easy, safe and promising management option for correcting congenital cervical and vaginal atresia.

Introduction

Congenital cervical atresia is a rare Müllerian anomaly and is associated with vaginal aplasia in 50% of cases. The true incidence of congenital cervical atresia is somewhat difficult to determine. Patients with congenital cervical atresia usually have a normal or deformed but functional uterus and present with cyclic abdominal pain with no menstruation. A timely surgical correction is necessary to relieve obstruction and to prevent the onset of endometriosis or hematometra.

Hysterectomy had been advocated by some clinicians for these patients due to risks of reoperation and potential death from reconstructive surgery. While hysterectomy resulted in irreversible physical and psychological damage for these teenage girls. With the development of new techniques and assisted reproductive techniques, fertility-sparing strategies, including uterovaginal or uterovestibular anastomosis, cervicovaginal canalization, cervicoplasty and cervical end-to-end anastomosis, had yielded encouraging outcomes, as well as successful pregnancies. But most of them were case reports and only in a short term follow up.

In our center, we applied laparoscopically assisted cervicovaginal canalization and vaginoplasty for patients with congenital cervical and vaginal atresia from Jan 2016. In this prospective observational study, we aimed to report the surgical procedure step by step and long-term outcomes of laparoscopically assisted cervicovaginal canalization and vaginoplasty for congenital cervical and vaginal atresia.

Materials And Methods

Setting and patients

Between January 2016 to July 2020, we carried out a prospective observational study of patients with congenital cervical anomalies who had received surgical treatment at the Department of Obstetrics and Gynecology, Peking Union Medical College Hospital (PUMCH).

The inclusion criteria were as follows. 1. Patients diagnosed with congenital cervical anomaly with a functional uterus, with or without vaginal atresia. The diagnosis of cervical atresia was suggested by clinical manifestation, gynecological examination, and results from pelvic Magnetic Resonance Imaging (MRI). In all cases, the diagnosis was confirmed during laparoscopy. 2. Patients had undergone laparoscopically assisted cervical canalization and vaginoplasty by the same experienced gynecologist (Dr. Lan Zhu). 3. Patients had undergone pelvic MRI examinations preoperatively.

The exclusion criteria were as follows: 1. The patient was diagnosed with distal vaginal atresia or transverse vaginal septum although the hematoma was present; 2. The canulations were not assisted by laparoscopy; 3. Patients with congenital cervical atresia were performed hysterectomy.

Data Collection

Clinical characteristics, including onset age, course of disease, malformations and lesions of the reproductive system, complicated malformations of other systems, and perioperative data, including operating time, estimated bleeding volume, intraoperative complications and postoperative febrile morbidity, were reviewed and recorded in detail. There is no widely accepted standard definition of febrile morbidity. We chose a level of 38°C, which has been used previously in a number of studies assessing postoperative infectious morbidity. Postoperative febrile morbidity was defined by a body temperature of at least 38 °C on two consecutive occasions at least 6 h apart, excluding the first 24 h.

Patients were followed up in outpatient clinic at 3, 6 and 12 months postoperatively, and once a year thereafter. Regarding menstruation, each patient was asked about her cycles and degree of dysmenorrhea. Gynecological examination was performed to measure vaginal length and to examine whether there was
restenosis.

Sexual satisfaction was assessed with the female sexual function index (FSFI), the most commonly applied and valid tool for measuring sexual satisfaction9. Chinese version of the FSFI has been validated to be a reliable and valid questionnaire in Chinese women10. Based on epidemiological research of Chinese women, the cutoff score for the FSFI total score was 23.4511. Higher scores indicate better sexual function.

**Outcome variables**

Success rate of laparoscopically assisted cervical canalization and vaginoplasty, which was defined as no restenosis.

**Surgery**

The surgical procedures were performed during menstruation. 1. Briefly, the patient was placed in the lithotomy position under general anesthesia. 2. Laparoscopy was performed to explore the abdominopelvic cavity to identify evidence of endometriosis, pelvic adhesions, and uterine anomalies, especially cervical anomalies and the amount of cervical remnants (Fig. 1A). If necessary, appropriate surgery, such as ovarian cystectomy, adhesiolysis, salpingostomy or excision of pelvic endometriosis, was performed. 3. The procedure was switched to vaginal surgery. To perform vaginoplasty, an 8-cm-long, two-finger-wide canal was created by blunt dissection between the bladder and the rectum along the anatomical vaginal route. To protect the bladder and rectum from injury, a metal urinary drainage tube was inserted into the urethra and the bladder, and an index finger was placed into the anus and rectum. 4. Laparoscopically, we dissected the uterine fundus by using a monopolar needle to access the uterine cavity. After suctioning the hematocoele, a metal guide was placed in the uterine cavity to push the uterus inferiorly (Fig. 1B). 5. The uterus was pushed inferiorly by the guide. In the meantime, a thick needle was punctured carefully through the neovaginal region toward the mass until the metal lever was reached. After ensuring no damage, an incision was made at the puncture site. The metal guide was pulled out (Fig. 1C). 6. A rubber cervical catheter, cut from a Pezzer catheter, was dragged from the vaginal vault to the uterine cavity. The mushroom-like end of the Pezzer catheter was inserted into the cervix (Fig. 1D). Excising the anterior uterine wall (Fig. 1E) and dragging the mushroom-like end of the catheter into uterine cavity (Fig. 1F). 7. The anterior uterine wall was closed using 2 − 0 a grain stitches (Fig. 1G). 8. Switching to vaginal surgery, circular suturing of the cervix to form the vaginal portion of the cervix and interrupted sutures were placed to secure the cervical catheter to the cervix (Fig. 1H). 9. A metal urinary drainage tube was inserted into the urethra and bladder, and methylene blue was injected into the bladder to check any outflow of the blue dye to reexamine for damage to the urethra or bladder. 10. Finally, after surgery, a soft mold made of two sterile condoms filled with sterile gauze was placed in the neovaginal cavity for the first 7–14 days (Fig. 1I). Later, the soft mold was removed, and the patient started vaginal dilation with a silicone dilator (length: 10 cm, diameter: 3 cm), which was advised to be daily, with 24 hours of continuous wear for the first 6 postoperative months to prevent contraction of the neovagina. Six months after surgery, the patient was advised to wear the mold intermittently and to gradually shorten their time wearing mold. There is no guideline for how long cervical stents should be placed. In our patients, cervical stents were not removed until the patient intended to be pregnant. If cervical stenosis occurred, a new cervical stent would be placed during reoperation. The schemas of surgical maneuver are presented in Fig. 2.

**Statistical analysis**

Statistical analysis was performed using SPSS (version 20.0 for Windows). The mean ± SD or median (quartile) is presented for quantitative variables, and frequencies (%) are presented for qualitative variables.

**Ethics approval**

This study was approved by the Ethics Committee of the PUMCH (project: S-452). All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

**Results**

From January 2016 to June 2019, a total of ten patients with congenital cervical and vaginal atresia who underwent laparoscopically assisted cervicovaginal canalization in the Peking Union Medical College Hospital were recruited in our study.

**Clinical characteristics of the patients**

The mean (± SD) symptom onset age was 14.5 ± 3.0 years. The median (quartile) duration of symptom onset to canalization was 15.5 (5.7, 28.0) months. Uterine corpus and cervical malformations were delineated by a combination of pelvic MRI and laparoscopic exploration. Cervical obstruction anomalies were observed in 6 (60%) patients, cervical agenesis was found in 3 (31.6%) patients, and cervical fragmentation was noted in 1 (10%) patients. All patients had total vaginal atresia. A total of 20% of the patients (2/10) had uterine malformations, including one with complete septate uterus (U2bC4V4) and one with a partial bicornual uterus (U3aC4V4) according to the European Society of Human Reproduction and Embryology (ESHRE) and the European Society for Gynecological Endoscopy (ESGE) classification12. The other 8 were all U0C4V4. Urinary ultrasound and/or intravenous pyelography examinations were performed in all 10 patients. None of them was found to have urinary anomaly. Plain-film X-ray exams of the spine were also performed in these 10 patients, one of them (1/10, 10%) was verified scoliosis (Table I).
Pelvic endometriosis was confirmed by pathological examination and stage of endometriosis was diagnosed by laparoscopy according to revised American Society for Reproductive Medicine classification of endometriosis. 20% of the patients had no endometriosis, 26% of the patients were diagnosed as stage I endometriosis, 30% of the patients were stage II and the remaining 30% were staged IV (Table I).

Surgical outcomes and follow-ups of the patients

All patients underwent laparoscopically assisted canalization smoothly, with no case requiring conversion to laparotomy and no intraoperative complications occurred. The mean operation time was 105.0 ± 34.4 min, and the median (quartile) estimated blood loss was 100.0 (87.5, 100.0)ml. The mean length of the neo-vagina was 8.3 ± 0.7 cm intraoperatively. The postoperative course was uneventful; only one case had a postoperative temperature of 38–39°C for 2 days after surgery.

The median (quartile) follow-up time was 26.0 (21.3, 48.3) months. All patients experienced relief of abdominal pain and resumed menstruation, including 9 patients with regular monthly menstruation. Eight patients experienced mild-to-moderate dysmenorrhea; the remaining 2 patients had no dysmenorrhea. During the follow-up period, cervical reobstruction occurred in one patient 12 months postoperatively, and ultrasound-guided translavaginal cervical dilation was performed. So far, eight months after the second surgery, no restenosis has been found.

The mean postoperative vaginal length was 7.9 ± 1.3 cm by the time of last follow up. 8 patients wore vaginal mold almost every day except for menstruation and 1 patient kept the length and width of vagina by regular sexual intercourse. The remaining 1 patient stopped dilating vagina 3 months after surgery. She had no vaginal restenosis and the vagina was about 7 cm long, but the vagina was only about 2 cm wide. Only one patient had intercourse sexual life. She was satisfied with sexual life subjectively. In addition, the scores of desire, arousal, orgasm, lubrication, satisfaction and pain were 6, 15, 16, 10, 12 and 11 respectively. After the weighted, the total FSFI score was 26.1. Moreover, this was the only patient prepared for pregnancy for two years, but she had not conceived yet (Table II). The patient is going to seek assisted reproductive technology for help.

Discussion

Congenital cervical atresia is a very uncommon Müllerian duct malformation. Timely surgical correction is necessary to prevent the development of endometriosis or hematosalpinx. The presence of vaginal atresia increases the difficulty of the surgical technique and leads to a higher rate of complications. Hence, the fertility-sparing procedure is a huge challenge for gynecologists when addressing these patients. Fujimoto et al. reported that the chance of success for canalization was approximately 40%-70% with or without vaginal atresia. In this prospective observational study, we showed an easy, safe and promising laparoscopically approach to cervical canalization in patients with congenital cervical and vaginal atresia. After 16–51 months of follow-up, the success rate of laparoscopic assisted cervicovaginal canalization was 90%.

Laparoscopic cervicovaginal canalization for congenital cervical malformations had been reported by many authors, most of them were case reports. Less invasive way for management of cervicovaginal aplasia, such as combined retropubic balloon vaginoplasty and laparoscopic canalization, had been reported in 4 cases with middle-term effective. While the complicated manipulation and risk of puncture made it difficult to be commanded. Laparoscopic cervicovaginal canalization with graft, such as full thickness skin graft and acellular porcine small intestinal submucosa graft, had also been reported to be effective in the midterm. In our cohort, we didn't use any graft to avoid additional damage or great expense.

There are some differences between our surgical technique and those proposed by the other authors. Our encouraging outcomes benefited from our attachment of great importance to protecting the bladder and rectum from injury, which we assumed would favor the success of the surgery. First, a metal drainage tube and finger were inserted into the bladder and rectum to indicate and protect these organs from being punctured or incised. Second, unlike previous reports, we didn't puncture from the fundus of uterus, placed a metal guide laparoscopically into the uterine cavity and then punctured from the vaginal vault to the uterus only when a mass was touched from the neovaginal vault, which avoided injuries caused by blind puncture. Third, unlike uterovaginal anastomosis, extensive dissection between the bladder and rectum was not necessary in our surgery, which therefore dramatically decreased the risk of injuries and pelvic adhesion and the risk of retrograde infection due to direct anastomosis with the endometrium.

Carefully outlining the anatomical variations of congenital cervical anomalies before performing any reconstructive procedure is recommended and crucial for prognosis. Cervical anomalies are classified into the following four types: 1) cervical agenesis: absence of the cervix; 2) cervical fibrous cord: a cervical body consisting of a fibrous band or cord; 3) cervical fragmentation: portions of the cervix are noted with no connection to the uterine body; and 4) cervical obstruction: an intact cervical body with obstruction of the cervical os. While no standards have been recommended for specific cervical anomalies. In our study, we attached great importance to preoperative and intraoperative evaluation of cervical malformations. But due to the small sample size, we couldn't figure out which cervical anomaly was good candidate for laparoscopically cervicovaginal canalization. Future studies are needed to address this problem.

A cervical stent seems to be necessary to drainage and to avoid cervical reobstruction. While retrograde infection should not be overlooked due to two cases of deadly peritonitis had been reported. According to our experience, Pezzer catheter should be given priority as cervical stent. Perforated, bulged “mushroom head” of Pezzer can drain menstrual blood and protect the stent from falling off.

Reproductive capacity and sexual satisfaction are useful detailed clinical information on treatment outcomes. Very few studies focused on that. Because in most cases, the congenital cervical atresia happened at the onset of puberty. Even after ten years of follow-up, the patients were only in their early 20 s. During this period, some patients underwent hysterectomies due to recurrent atresia or severe complications; the remaining patients were too young to be pregnant. Successful pregnancy after assisted reproductive techniques had been reported in several cases. In our cohort, only one patient had intercourse sexual
life. Encouragingly, she felt satisfied with sexual life and got along with her partner. But she hadn't been conceived after two-year preparation. The strictured cervix, pelvic inflammatory environment and other factors could result in the infertility. Future studies are needed to discuss the pregnant rate because of its important clinical meaning and practical value.

**Strengths and limitations**

In this study, we described the surgical procedure step by step and used surgical photos and schematic to show the surgical procedure more intuitively. In addition, our cohort had a long follow-up term, with a median follow-up duration of 26 months. The small sample size was the primary limitation of our study, more patients need to be enrolled in a future study.

**Conclusion**

In summary, in this prospective observational study, we reported that laparoscopically assisted uterovaginal canalization and vaginoplasty is an easy, safe and effective management option for relieving obstructions in patients with congenital cervical and vaginal atresia. However, more studies and longer follow-up are needed.

**Declarations**

**Ethics approval and consent to participate:**

This study was approved by the Ethics Committee of the PUMCH (project: S-452).

**Consent for publication:**

Written informed consent for publication was obtained.

**Availability of data and materials:**

All data generated or analyzed during this study are included in this published article.

**Competing interests:**

The authors declare that they have no competing interests.

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**Authors' contributions:**

J.K. participated in the design of the study, conducted the data acquisition, interpreted and analyzed the data, and drafted and revised the manuscript. C.N. pointed out deficiencies and ameliorated the manuscript. Y.Z participated in videoing the surgery and selecting of photographs. C.C.M., Y.D.M., Y.W. and W.J.T contributed to the data collection. L.Z. was responsible for design of the work and final approval of the version to be published.

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| Patient No. | 1   | 2   | 3   | 4   | 5   | 6   | 7   | 8   | 9   | 10  |
|------------|-----|-----|-----|-----|-----|-----|-----|-----|-----|-----|
| Age at diagnosis | 13  | 14  | 17  | 14  | 19  | 13  | 19  | 10  | 14  | 12  |
| Delay in diagnosis (month) | 24  | 16  | 24  | 15  | 72  | 12  | 40  | 5   | 6   | 1   |
| Uterine anomaly | Complete septate uterus | N | Partial bicorporeal uterus | N | N | N | N | N | N | N |
| Cervical anomaly | Cervical obstruction | Cervical obstruction | Cervical absence | Cervical obstruction | Cervical Fragmentation | Cervical absence | Cervical absence | Cervical obstruction | Cervical obstruction | Cervical obstruction |
| Preoperative vaginal length (cm) | 0   | 0   | 1   | 0   | 0   | 0   | 2   | 0   | 0   | 0   |
| Renal anomaly | N   | N   | N   | N   | N   | N   | N   | N   | N   | N   |
| Spinal anomaly | N   | N   | N   | N   | N   | N   | Scoliosis | N   | N   | N   |
| Preoperative AFS stage* | 0   | I   | IV  | II  | II  | IV  | II  | IV  | I   | 0   |
| Previous surgery | 0   | 0   | 1   | 0   | 0   | 0   | 0   | 0   | 1   | 0   |

N: normal;
*: Diagnosed by laparoscopy.
Table II
Surgical outcomes of the 10 patients

| Patient No. | 1  | 2  | 3  | 4  | 5  | 6  | 7  | 8  | 9  | 10 |
|-------------|----|----|----|----|----|----|----|----|----|----|
| Surgical time (min) | 80 | 90 | 80 | 120| 120| 180| 60 | 120| 80 | 120|
| Estimated blood loss(ml) | 50 | 100| 100| 100| 100| 300| 100| 100| 10 | 100|
| Intraoperative complications | NO | NO | NO | NO | NO | NO | NO | NO | NO | NO |
| Vaginal length at surgery | 8  | 8  | 8  | 8  | 8  | 8  | 10 | 8  | 9  | 8  |
| Postoperative morbidity | NO | NO | NO | NO | NO | NO | NO | Yes| NO | NO |
| Follow-ups (Months) | 22 | 19 | 27 | 49 | 30 | 48 | 16 | 22 | 25 | 51 |
| Stricture of the cervix | NO | Yes| NO | NO | NO | NO | NO | NO | NO | NO |
| Menstruation | Regular, 2-3/23 | Regular, 7/28-29 | Regular, 5-7/30 | Regular, 3/23-24 | Irregular | Regular, 5-6/30 | Regular, 4-8/30 | Regular, 10/23-30 | Regular, 7/35-37 | Regular, 4-5/30 |
| Dysmenorrhea | Mild | Moderate | NO | Mild | Mild | NO | Moderate | Moderate | Mild | Mild |
| Vaginal length at the last follow-up(cm) | 7  | 7  | 8  | 10 | 8  | 8  | 10 | 6  | 8  | 7  |
| Vaginal dilation | By mold, every day | By mold, every day | Regular sexual intercourse | By mold, every day | By mold, every day | By mold, intermittently | By mold, every day | By mold, every day | Stop dilating 3 months after surgery |
| Sexual satisfaction | NK* | NK | Yes | NK | NK | NK | NK | NK | NK | NK |
| Pregnancy | NO | NO | Prepared for 2 years, haven't pregnant | NO | NO | NO | NO | NO | NO | NO |

*NK: Not known, because of no intercourse sexual life.

Figures
Figure 1

Intraoperative images of laparoscopically uterovaginal canalization. (A) Laparoscopically images of pelvic cavity. The uterus was enlarged with no anomaly. No evidence of pelvic endometriosis or adhesion was found in this patient. (B) A metal guide was placed in the uterine cavity to push the uterus inferiorly. (C) The metal guide was pulled out from uterine cavity to the neovagina. (D) The mushroom-like end of the Pezzer catheter was inserted into the cervix. (E) Excising the anterior uterine wall. (F) Dragging the mushroom-like end of the catheter into uterine cavity. (G) Closing the anterior uterine wall. (H) Interruptedly suturing the cervical catheter to the cervix. (I) A soft mold made of two sterile condoms filled with sterile gauze was placed in the neovaginal cavity.

Figure 2

Surgical maneuver schemas. (A) Prior to the operation, the uterine cavity was enlarged due to hematoma, the uterine cervix was absent and the vagina was unobservable. (B) The neovagina was manually constructed between the bladder and the rectum and toward the uterus. To protect the bladder and rectum
from injury, a metal urinary drainage tube was inserted into the urethra and the bladder, and an index finger was placed into the anus and rectum. (C) The uterus was pushed inferiorly by the metal guide. (D) The cervix was sutured with the neovagina, and the cervical catheter was interrupted sutured to the cervix.