

Successful Laparoscopic Sigmoid Transposition for Cervicovaginal Agenesis in Presence of Functional Uterus; a Case Series

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Abstract

Background

This study has been designed to investigate the long-term outcomes of laparoscopic sigmoid cervicovaginoplasty in patients with congenital cervicovaginal atresia with a functional uterus and stepwise description of surgery.

Methods

Seven patients with cervicovaginal atresia with a functional uterus, underwent laparoscopic sigmoid cervicovaginoplasty surgery, between 2016 and 2020. All subjects underwent a successful surgery with no major complications. Mean follow-up duration was 25.9 months (2-48 months). All the patients had regular menstrual cycles. The average length of vagina was 8.9 cm (7.6-10.5 cm). In one patient, proximal stenosis of neovagina was observed 12 months after surgery.

Results

The mean age and BMI of the patients were 18y (12-27y) and 19.7 (17.6-22.4). The average time period between the initiation of disease symptoms and the operation was 52.28 months (2-156 months). We had no significant post-operative short term complication. We did not perform hysterectomy in our patients either at the time of first surgery or as a result of a complication. None of our patients complained of signs of low anterior resection syndrome. Two subjects had sexual activities without dyspareunia, post coital bleeding, or malodor vaginal discharge.

Conclusion

Sigmoid cervicovaginoplasty is a safe and effective procedure with satisfactory long term outcomes. This surgery eliminates the psychological burden of hysterectomy in these patients. Through preserving the uterus, patients may have a chance of possible future pregnancy if abdominal cerclage is performed.

Background

Congenital cervical agenesis is a female genital tract anomaly with an incidence of 1:80000 – 1:100000 [1], and approximately half of the cases suffer from congenital vaginal agenesis [2]. Less than 200 cases of cervicovaginal agenesis have been reported since 1942. The incidence is far less if it coexists with functional uterus [3].

This condition usually presents with cyclic abdominal pain and amenorrhea, and if not diagnosed early, could give rise to hematometra and endometriosis as a result [4].

Different surgical approaches have been suggested to create neocervix and neovagina in order to drain blood from the uterus; however, considering the complications and risks of stenosis and second surgery, hysterectomy used to be considered as the treatment of choice [5]. Cervicovaginal agenesis management is performed to diminish the symptoms of cervicovaginal obstruction, achieve appropriate sexual function, and preserve the uterus for future pregnancies [6].

The main objective of creating neovagina is to create a canal with an appropriate length, diameter, and axis and normal lubrication for a desirable sexual intercourse [7, 8]. Using rectosigmoid to make neovagina is among the new methods of creating neovagina [9–11]. Sigmoid vaginoplasty using laparoscopic technique was first described by Ohashi in 1996 [12]. The advantages of rectosigmoid transposition are its similarities with normal vagina in length, texture, and appearance. Moreover, it could simulate a vaginal lining with natural lubrication [13].

This study is the first report of laparoscopic sigmoid cervicovaginoplasty in seven patients with functional uterus.

Methods

In this study, sigmoid cervicovaginoplasty was performed between 2016 and 2021 in seven patients with cervicovaginal agenesis and functional uterus. The study was done in a private hospital and a university hospitals affiliated with Shiraz University of Medical Sciences (SUMS), Shiraz, Iran. This study was revised and approved by the Ethics Committee of Shiraz University of Medical Sciences (IR.SUMS.REC.1400.034). All the surgeries were done by the first author (SA).

The average age of the subjects was 18 years (12y-27y). All of the patients suffered from cyclic abdominal pain and primary amenorrhea. No vagina and cervix were seen upon physical examination and the uteruses were larger than normal. Transrectal ultrasound with bowel preparation and MRI revealed cervicovaginal agenesis along with hematometra in all subjects. Endometriosis was confirmed in two patients (18 and 27y).

Preoperative consultation was done for each patient. The patients and their guardians were informed about the details of surgical procedure and its possible complications, including fistula, anastomosis leakage, peritonitis, diversion colitis, bowel obstruction, introital stenosis, constipation, mucocele, and excessive and malodor vaginal discharge. The patients were further informed about alternative surgical procedures, such as cervicovaginoplasty using peritoneal graft, cervical canalization, and split and full thickness skin graft [4, 6, 14]. Due to social and cultural limitations, lack of uterus may have adverse effects on patient's life. Therefore, hysterectomy was not a first line treatment option for our patients. We suggested the use of pressure on perineum to make a pouch before surgery but due to the same cultural beliefs (prohibition of sexual activity before marriage), our patients refused it. We explained to patients that in the case of failure there is a possibility of hysterectomy, and written informed consents were obtained from all the patients or their legal guardians if they were underaged. Moreover, in younger patients, the patient and her legal guardians (parents) were suggested to postpone surgery by suppressive medical therapy until psychological consultation was completed and the patient was ready for the procedure [6]. Finally, the patient was informed that abdominal cerclage as a complementary procedure might be required if she wanted to get pregnant in the future. The subjects were then followed up for at least one year, except the sixth patient as the last surgery was done just 2 months prior to this article.

Surgery

After the intravenous administration of antibiotic and urinary catheterization, laparoscopy was performed under general anesthesia in low lithotomy position. Initially, a 10 mm port was inserted through the umbilicus, except for one subject with hematometra and 28 w uterus in which the primary port was inserted through a surface between xiphoid and umbilicus, and the peritoneal cavity was insufflated by carbon dioxide.

Afterwards, four ancillary ports were placed (5 mm in right and left iliac fossa, left paraumbilicus, and 10 mm in suprapubic) under direct vision. The abdominopelvic cavity was explored for the possible existence of endometriosis

or pelvic adhesion and to determine the type of anomaly in uterus and cervix. Endometriosis was diagnosed in two patients and adhesiolysis and resection of pelvic and ovarian endometriosis were done simultaneously.

Following the identification of the ureters, dissection of pararectal space, and release of sigmoid from the left lateral pelvic sidewall, a 12-15 cm segment of distal sigmoid was selected and mobilized while appropriate vasculature and sigmoidal artery were preserved. The suprapubic port was changed to 12 mm. The proximal and distal parts of the harvested segment of sigmoid were cut using two EndoGIA (size 60) purple staplers (Covidien, Norwalk, CT, USA).

A suprapubic incision (3 cm) was performed and after placing the Alexis retractor (Applied medical, Rancho Santa Margarita, CA, USA), the distal segment of the descending colon was pulled out through incision and an anvil was inserted into it. The continuity of the colon was restored and the anastomosis between the distal segment of the descending colon and rectum was completed by EEA circular stapler number 29 or 32 (Covidien, Norwalk, CT, USA). Subsequently, the safety of the anastomosis was confirmed using flat air leakage test.

In the next step, the procedure was switched to a vaginal approach. With a great concern for bladder and rectal protection, after making a transverse incision in the appropriate place on perineum, a two-finger-width canal was established in vesicorectal septum up to the posterior cul-de-sac by placing a bladder retractor in the anterior and putting a finger inside the rectum.

Under laparoscopic vision and with the guidance of a manipulator, posterior cul-de-sac was opened by monopolar hook. The harvested sigmoid segment was moved into the dissected place in isoprismatic direction, and the distal end of the segment was fixed to perineum with eight seromuscular sutures (Vicryl 2/0, Ethicon, Somerville, NJ, USA). Next, an elliptical incision was made in the distal posterior lower segment of the uterine myometrium. After drainage of the hematometra and irrigation of the uterine cavity, the proximal end of the harvested segment of sigmoid was connected to the uterus. Seromuscular sutures were performed in a separate fashion using vicryl 2/0 (Ethicon, Somerville, NJ, USA). Finally, intraperitoneal drain (Jackson-Pratt drain, Fushan Medical, Shanghai, China) and anti-adhesive barrier film (Surgiwrap, MAST Biosurgery, San Diego, USA) were placed in the posterior cul-de-sac (figures 1, 2).

Postoperative care

The patients were discharged after a minimum of six postoperative days. They were ambulated two days after surgery. Foley catheter was removed on the third postoperative day. Diet started on the 5th postoperative day. Neovagina was examined daily and the patient was monitored for any complication of surgery such as anastomosis leakage. We did not suggest wearing a vaginal mold to any patient.

Postoperative follow-up protocol

All of the patients underwent examination one week and then one, three, six, and twelve months after surgery, and then annually.

The follow-up included the assessment of vaginal health and the function of genital system, including the presence of dysmenorrhea, regularity of menstrual cycle, and abnormal vaginal discharge. Sexually active subjects were questioned about dyspareunia and sexual satisfaction using FSFI questionnaire (female sexual function index). Ultrasound screening was performed for any sign of hematometra, pelvic endometriosis, and pelvic abscess.

In sexually active patients, sexual intercourse was permitted two months after operation.

Results

Patient demographics

At the time of operation, the mean age and BMI of the patients were 18y (12-27y) and 19.7 (17.6-22.4), respectively. The average time period between the initiation of disease symptoms and the operation was 52.28 months (2-156 months).

All of the patients had complete vaginal agenesis with functional uterus. Two patients had vaginal dimples of about 1-2 cm. Complete cervical agenesis was diagnosed in three patients, one had fibrotic band and the rest had cervical fragmentation. Normal uterus was found in four patients ($U_0C_4V_4$). Bicornual septate uterus was diagnosed in one patient ($U_{3C}C_4V_4$). One subject had unicornuate uterus with a non-functional, non-communicating rudimentary horn ($U_{4a}C_4V_4$) and the seventh one had bicornuate uterus ($U_{3a}C_4V_4$) according to European Society of Human Reproduction and Embryology (ESHRE) and the European Society for Gynecological Endoscopy (ESGE) consensus on the classification of female genital anomalies [15]. Just one patient with unicornuate uterus had one kidney, and musculoskeletal anomalies were not observed in any of the participants.

Two of the patients had a history of failed cervicovaginoplasty with peritoneal graft and amnion graft. Endometriosis was diagnosed in two patients, which was confirmed by histopathological findings (Table 1).

Table 1
Patient demographics

Case	1	2	3	4	5	6	7
Age	12	27	27	18	15	13	14
Uterine anomaly	Bicornual septate	Normal	Normal	Normal	Normal	Unicornuate with rudimentary horn	Bicornuate unicornis
Cervical anomaly	Fragmented cervix	absent	absent	absent	Fragmented cervix	Fragmented cervix	not-canalized cervix
Preop vaginal length	Absent	absent	absent	2 cm	Absent	absent	3 cm
Delay in diagnosis	2 months	156 Months	144 months	48 months	12 months	2 months	2 months
BMI	18.7	22.4	18.6	20.3	22.3	17.6	18.1
History of cervicovaginoplasty	Yes Peritoneal graft	No	No	No	No	No	Yes Mc Indoe with amnion graft
Other anomalies	No	NO	No	NO	No	Absent left kidney	No

Surgical characteristics and intraoperative complications

The average time of the operation was 203.5 minutes and was longer in patients with endometriosis. All surgeries were done laparoscopically and without the need for conversion to laparotomy. In one patient, inadvertent rectal injury was observed, which was diagnosed and repaired immediately at the time of surgery. The average intra-operative blood loss was 250 ml and none of the patients required blood transfusion. No significant post-operative short term complication, such as bleeding, surgical site infection, hematoma, anastomosis leakage, urinary complications, fistula formation, and mucosal prolapse was reported. In one subject, low grade fever was recorded on the 2nd and 3rd postoperative days, which was subsequently resolved with conservative management.

Long-term post-operative

Mean post-operative follow-up time was 25.9 months (2-48 months). One subject has only been followed for two months and we therefore no long term outcome data for her. During the follow-up period, each of the first six patients had regular menstrual cycles without any abnormal uterine bleeding (AUB). In the first three months following the surgery, three patients experienced heavy menstrual bleeding which was resolved without any intervention.

One patient had moderate dysmenorrhea (without any sign of endometriosis intra or post-operative) but others reported no pain.

Two patients reported copious malodorous vaginal discharge in the first three months after the procedure, which was diminished instantaneously.

None of our patients complained of signs of low anterior resection syndrome (LARS) such as bladder atonia and constipation.

We had no mucosal prolapse or introital stenosis among our patients. No recurrence or new onset endometriosis was observed in the post-operative follow-up of the patients.

The average length of neovagina was 8.9 cm (7.6 to 10.5 cm).

Two patients had satisfied vaginal intercourse without any dyspareunia, post coital bleeding, or malodor vaginal discharge. These two patients had not tried to conceive before the study.

A year after the sigmoid cervicovaginoplasty, one of our patients developed stenosis of the proximal part of neovagina. In vaginoscopy, the proximal part was fibrotic and obstructed with adequate vascularized tissue. Therefore, resection of stenotic segment and reanastomosis of the neovagina to the uterus were performed by laparotomy. The mentioned patient also suffered an episode of PID and pyometra 14 months after the second surgery, which was treated conservatively and now she has regular menstrual cycles without any other symptoms (Table 2).

We did not perform hysterectomy in our patients either at the time of first surgery or as a result of a complication.

Table 2
Intraoperative and postoperative findings

Case	1	2	3	4	5	6	7
Operation time	185	250	155	285	210	160	180
Bleeding	250	450	150	350	200	150	200
Endometriosis	No	No	Score=48 Satge IV Bilateral hematosalpinx, Bilateral Uterosacral ligament DIE 4 points of superficial endometriosis	Score=36 Stage III Left endometrioma and left hematosalpinx	No	No	No
Intraoperative complication	No	No	No	Accidental perforation of bowel	No	No	No
Post op follow up period	18 months	35 months	39 months	35 months	48 months	4.5 months	2 months
Short term complication	Low grade fever	No	No	No	No	No	No
Long term complication	No	No	No	No	Proximal stenosis/PID	No	NK
menstruation	Regular (3 month HMB**)	Regular (3 month HMB)	Regular	Regular (3 month HMB)	Regular	Regular	NK
Vaginal length	8	9.5	8.5	8.2	7.6	10.5	10
dysmenorrhea	VAS 5/10	No	No	No	No	No	NK
Dyspareunia	NK	No	NK	No	NK	NK	NK
FSFI score***	NK	23	NK	26	NK	NK	NK
Vaginal discharge	Yes, just 3 months	No	Yes, just 3 months	No	No	No	No

* NK= Not known. ** HMB= Heavy menstrual bleeding. *** FSFI= Female Sexual function Index.

**** LARS=Low anterior resection syndrome.

Case	1	2	3	4	5	6	7
LARS****	No						
* NK= Not known. ** HMB= Heavy menstrual bleeding. *** FSFI= Female Sexual function Index.							
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Discussion

In the case of cervicovaginal agenesis and functional uterus, surgery consists of three steps: 1- creation of neovagina, 2-creation of neocervix, and 3-maintaining the continuity between neovagina, neocervix, and uterus [6].

However, when there is no sufficient cervical tissue, the outcome of cervicovaginal reconstruction remains unclear and there is a high probability of cervical stenosis that may lead to hysterectomy [16].

Bowel vaginoplasty was traditionally used in transgender women or biologic women with failed skin graft vaginoplasty [17]. It is a kind of direct uterovaginal anastomosis which can be used for any kind of cervical anomaly (fibrotic band, fragmentation, complete absence) if performed by our technique, in which cervix is bypassed.

Baldwin did the first bowel vaginoplasty in 1907 with a segment of ileum and in 1911, Wallace performed sigmoid colon vaginoplasty [18]. Finally, Ohashi reported laparoscopic sigmoid vaginoplasty for the first time in 1996 [12].

Different segments of bowel have been used for vaginoplasty, such as sigmoid, ileum, cecum, and ascending colon. But the best alternative is sigmoid which has an appropriate diameter, produces lubricant mucosa, and due to its location, has the best accessibility for anastomosis as a neovaginal conduit [19].

Bowel vaginoplasty is a practical choice at any age because it creates a conduit with potential growth. Sigmoid neovagina is resistant to mucosal injury with a very low incidence of graft necrosis because of preservation of its vascularity [19].

In 2011, Kim et al. reported that 6 and 12 months post operation, sigmoid neovaginal pH was 6.5 and 6, and squamous transformation of columnar epithelium was observed in only one of the 12 patients [20].

Predisposing factors for bowel-related complications in sigmoid vaginoplasty are tension over vascularized pedicle, short mesentery, obesity, and masculinized pelvis. So, surgeons should be highly skilled [21].

We had a very small intraoperative complication only in one patient whose rectum was inadvertently opened which was diagnosed and repaired immediately. Risk of rectovaginal fistula in primary cervicovaginoplasty is about 0-4%. But we had no rectovaginal or ureterovaginal fistula [17].

In the literature, the risk of anastomosis leakage is 0-7.1% and we had no anastomosis leakage in our series [22].

We had no post-operative ileus and stenosis of anastomosis site.

Bouman et al. performed 42 transgender sigmoid vaginoplasties and reported one (2.4%) case of anastomosis leakage, six (14.6%) patients with introital stenosis, and one mucosal prolapse subject. We had no mucosal prolapse or introital stenosis in our study [17].

None of our patients needed blood transfusion, and they had no post-operative surgical site infection.

In the literature, neovaginal stenosis is divided to two subtypes: introital stenosis (0-55.6%) and diffuse stenosis [17]. One of our patients had only proximal part stenosis and an episode of PID and pyometra 14 months after the second surgery. Due to this episode of infection, there was a possibility of stenosis and incomplete drainage. However, she refused vaginoscopy and further investigations. But she has a regular period.

In 2009, Kannaiyan et al. reported a series of 11 subjects among whom seven patients had cervicovaginal agenesis and functional uterus and underwent sigmoid vaginoplasty by open method. The mean operation time was 239 min, they reported that no mold was required to prevent stenosis and only one (9%) patient had introital stenosis. They reported no other complications [23]. Our method is similar to theirs, but it was done by laparoscopy and the mean surgical time was lower in our study (207 min) despite the laparoscopic route.

The important point in these patients is to build a wide and patent neovagina-uterus anastomosis, to completely drain the uterus and diminish the risk of pyometra as a result of incomplete uterus drainage and the recurrence of dysmenorrhea and endometriosis [23]. In agreement with kannaiyan et al., in our study, resection of a healthy portion of myometrium was required to maintain an open standing anastomosis to reduce the risk of ascending infection [23]. By this method, we are able to make a non-competent free drainage passage. Therefore, if the patient desires future pregnancy, she needs abdominal cerclage before attempting to get pregnant. We did not put abdominal cerclage simultaneously with cervicovaginoplasty.

Additionally, we should caution the patients against natural vaginal delivery and emphasize the necessity of cesarean section [23].

Kisku et al. reported 20 patients with cervicovaginal atresia, functional uterus and open colovaginoplasty among whom the rate of anastomosis leakage and recurrence of dysmenorrhea and endometriosis was 5% and 10%. Mild stenosis of neovagina were detected in 10% of the patients [24].

Yang et al. reported 22 cases of laparoscopic assisted sigmoid vaginoplasty. But they performed hysterectomy for all patients with functional uterus (15 patients) due to cervical agenesis. In their study, the mean time of surgery was 279 ± 30 min, the mean estimated blood loss was 334 ± 71 cc and the mean neovaginal length was 11.3 ± 1.2 cm [25].

The mean operation time in our study was 203.5 min, which is shorter than Kannaiyan series (laparotomy, 239 min) and Yang series (279 min, laparoscopic vaginoplasty and hysterectomy) and similar to Bouman series (210 min, laparoscopy in transgender women) despite the fact that we had an additional procedure of uteroneovaginal anastomosis in our patients [17, 23].

In our patients, the mean vaginal length was 8.9 cm (7.6-10.5) which is a very good length.

(Bouman: 16.3 ± 1.5 cm, Cao: 12.5cm in and Yang: 11.3 ± 1.2 cm) [17, 19, 25].

Given that cervicovaginal agenesis is mostly diagnosed and treated at peripubertal age, even after long follow-up periods, patients may not reach the age to attempt to get pregnant. Especially in our society, with cultural limitations, women may not have a sexually active life prior to matrimony.

In our study, just two patients were sexually active and both were satisfied with their sexual relationship. None of them had attempted to get pregnant prior to the research.

Due to rarity of this congenital malformation, we had a small sample size so the low rate of adverse events is not reliable in our study.

Although all the surgeries in our center were done by a gynecologist who is skilled in advance minimally invasive surgery (SA), it can also be done by multidisciplinary approach and collaboration of gynecologists and colorectal surgeons.

Conclusion

Sigmoid cervicovaginoplasty is a safe and effective procedure with satisfactory outcomes during long term follow-ups. This surgery eliminates the psychological burden of hysterectomy in these patients and if it is done by laparoscopy, patients will benefit from the advantages of minimally invasive procedures such as cosmetics (open surgery requires at least 12 to 15 cm of incision) [20], less adhesion formation, lower infection rate and wound complications, ileus, and accelerated recovery time [17]. By preserving the uterus, we can make future pregnancy possible by performing abdominal cerclage in these patients.

Our study is the first to perform the procedure by laparoscopy and have a follow-up of patients.

Abbreviations

HMB: Heavy menstrual bleeding, FSFI: Female sexual function index, LARS: Low anterior resection, syndrome.

Declarations

Ethics approval and consent to participate: This study was revised and approved by the Ethics Committee of Shiraz University of Medical Sciences (IR.SUMS.REC.1400.034). All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1964 and its later amendments. The written informed consents were obtained from all the patients or their legal guardians if they were underaged

Consent for publication: Not applicable.

Availability of data and materials: The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

Competing interests: The authors declare that they have no competing interests.

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Authors' contributions:

SA: Performed all the surgeries, EHN: Wrote the article and was an active team member at the time of surgeries, EA: Writing the article and was an active team member at the time of surgeries, SA: Wrote and edited the article, KC: Done the data collection part and was an active team member at the time of surgeries, ZZ: Done the data collection

part and patient follow-up, TP: Performed post-operative ultrasonography and patient follow-up. All authors read and approved the final manuscript.

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Figures

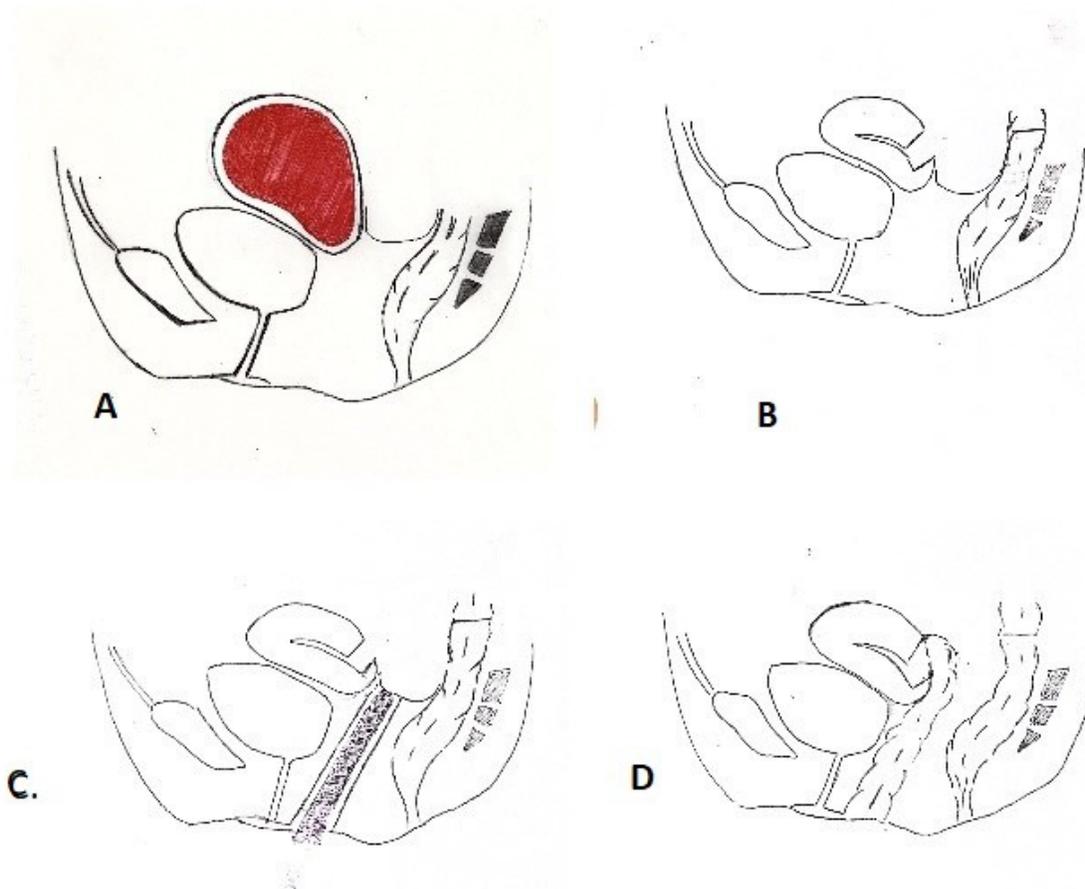


Figure 1

Diagrammatic figure of the procedure. (A) At the beginning of surgery, the uterine cavity was enlarged due to hematoma. The uterine cervix and the vagina were absent. (B) After separating a sigmoid segment and restoring the continuity of rectum, a wide elliptical window was made in the lower segment of the uterus. (C) The neovagina was manually constructed between the bladder and rectum and posterior culdesac was opened by the guide of a manipulator. (D) The harvested sigmoid segment was moved into the dissected place in isoprismatic direction, and the distal end of the segment was fixed to the perineum. Proximal end of the harvested segment of sigmoid was connected to the uterus.

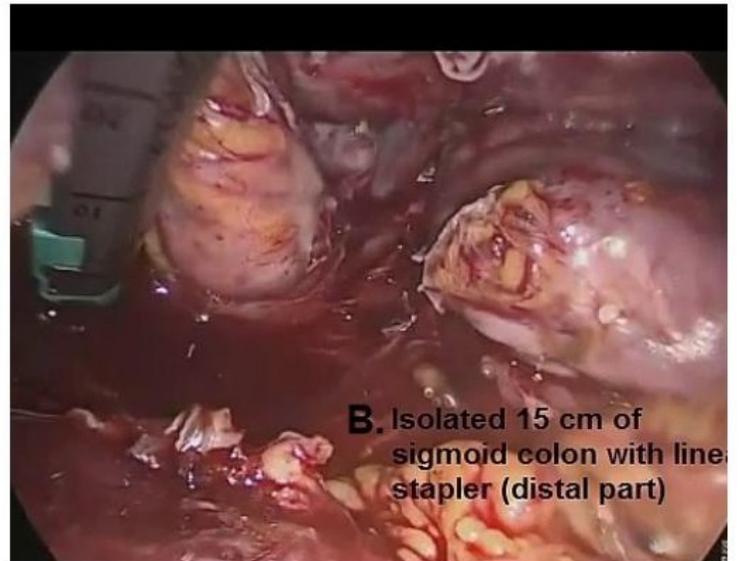


Figure 2

Procedure: stepwise