

An innovative and effective approach of cervical canal penetration in patients diagnosed with type II vaginal atresia with adequate uterine corpus development: A case series

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Abstract

We introduced our experience for an innovative and effective approach for cervical canal penetration in patients diagnosed with type II vaginal atresia with adequate uterine corpus development. The uterine cavity and neovagina was connected through the puncture set for percutaneous nephrostomy while maintaining the integrity of the uterus and cervix under laparoscopic and transvaginal ultrasound monitoring. A porcine small intestine submucosal graft was fitted into the artificial vaginal wall. There were few side effects and no obvious sequelae.

Keywords: Vaginal atresia; Congenital reproductive tract abnormality; Minimally invasive operations

1. Introduction

Congenital vaginal atresia is a rare congenital reproductive tract abnormality.^[1] Patients with type I vaginal atresia have a normally developed upper vaginal segment, cervix, uterine corpus, and functional endometrium. The symptoms are severe and appear earlier in patients with this disease. Type II vaginal atresia is often accompanied by cervical dysplasia, a functional endometrium, and a normally or abnormally developed uterine corpus.^[2] The main clinical manifestations are no menarche, periodic abdominal pain, or pelvic masses. Symptom severity is associated with endometrial function.^[3]

Type II vaginal atresia has various clinical manifestations,^[4] and its treatment should be individualized. Owing to the low success rate of the creation of neovaginas and cervicoplasty as well as the low rate of pregnancy, hysterectomy has been proposed as a therapeutic surgery for many years. To date, uterine preservation has not been recommended for patients with poor cervical or uterine corpus development. Vaginal reconstruction and cervicoplasty may be considered for patients with a better developed cervix and normal uterine corpus, as well as those without severe pelvic endometriosis.^[5] Penetration of the cervix and the uterine cavity (referred to as cervical canal penetration) is essential for uterine

preservation. There is no satisfactory approach for cervical canal penetration, and almost all surgical methods undermine the integrity of the cervix or uterine corpus and even require removal of the dysplastic cervix. Herein, we report 4 cases of type II vaginal atresia to explore an innovative approach, which was called laparoscopic and ultrasound-guided cervical canal penetration after vaginal reconstruction. It was used to treat patients with type II congenital vaginal atresia with adequate uterine corpus development.

2. Materials and methods

The study included 4 patients who had visited our hospital's gynecological department. All patients were diagnosed with type II vaginal atresia with adequate uterine corpus development and underwent laparoscopic and ultrasound-guided cervical canal penetration after vaginal reconstruction at our hospital. This study was approved by the Ethics Committee of our hospital. The patients and guardians were fully informed of the purpose of the surgical procedure and the possible complications before providing written consent to undergo the surgical procedure.

2.1. Surgical procedures

Physical examination revealed that all 4 patients had only a 1- to 2-cm blind end at the vaginal external opening with proximal occlusion, and the cervix could not be exposed. The patient received a clyster before surgery. During the surgical process, a catheter was placed to drain urine and indicate the position of the urethra. The assistant placed a finger in the rectum for guidance, and the vaginal hollow was incised at the 4- and 8-o'clock positions. After the incision of the vaginal hollow was made, the loose tissue was sequentially expanded with a cervical dilator to 11 mm from the cervix under transanal ultrasound monitoring. An artificial vaginal mediastinum was incised to form a new vaginal cavity with a length of approximately 9 cm and a width of approximately

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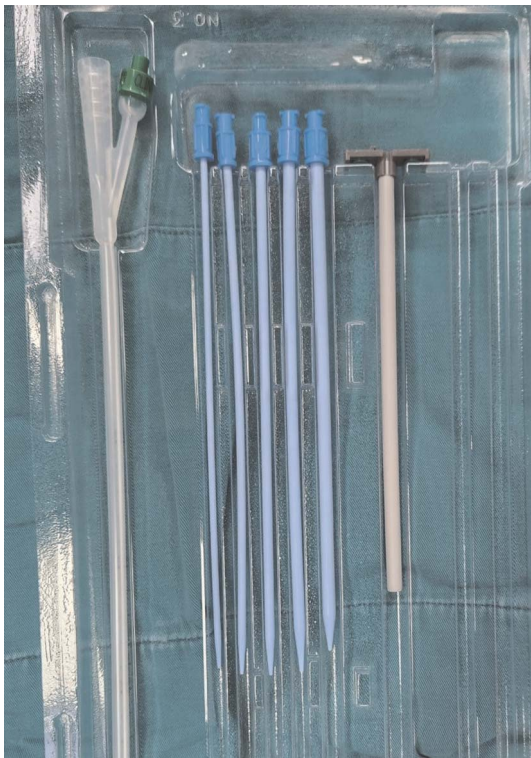


Figure 1. Puncture set for percutaneous nephrostomy.

4 cm. Under transvaginal ultrasound guidance, the atretic cervix was confirmed to be in line with the endometrial line and artificial vagina. The puncture needle was placed under the guidance of laparoscopic monitoring and transvaginal ultrasound, which accurately reached the uterine cavity through the atretic cervix and gradually expanded the tissues through the puncture set for percutaneous nephrostomy (Fig. 1). When it was difficult to insert the dilator, the retroperitoneal folds of the bladder and uterus were opened under laparoscopic guidance. The bladder was then pushed down, and the anterior and posterior fornixes were cut as needed so that the cervix could be clamped through the vagina and the uterus could be pulled down. A dilation rod was used to dilate the cervical canal, and a sterile Malecot catheter was inserted for drainage (Fig. 2). After the uterine cavity was drained, the anterior and posterior fornices were sutured. A porcine small intestine submucosal graft (Beijing Datsing Bio-Tech Co., Ltd., Beijing; model: 10 × 15 cm) was trimmed and put into the artificial vaginal wall. Vaginal mucosa fragments were placed between the small intestine submucosal graft and neovagina. The sleeve was then sutured to the reconstructed vaginal cavity. Finally, the iodophor gauze was placed in a sterile condom and stuffed into the neovagina. The labia minora was sutured to prevent the packed items from falling. The drainage tube was connected to a drainage bag, and the drainage volume was recorded daily after the operation.

2.2. Postoperative treatment

Vulvar disinfection was performed daily after surgery. An analgesic pump was used for 1 week before being replaced with oral analgesic tablets 1 week after surgery. Two weeks after the operation, the stitches of the labia minora and gauze wrapped with the original condom in the vagina were removed under intravenous general anesthesia and replaced by a special sterile tubular stent. All

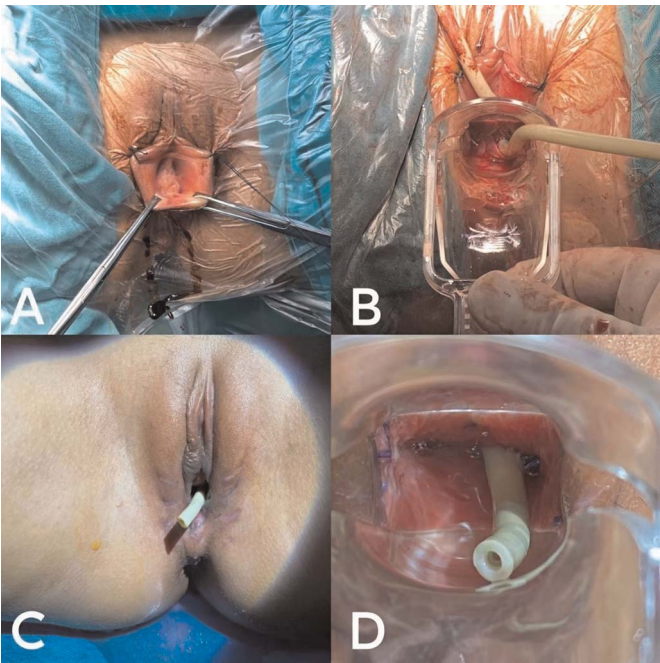


Figure 2. Photographs of 1 of the 4 patients with type II vaginal atresia before and after the surgical procedure. (A) Patient had vaginal atresia. (B) A Malecot catheter was inserted for drainage during the surgical procedure. (C and D) The vagina did not show infection or adhesion 3 weeks after the surgical procedure, and the drainage tube was in the correct position and unblocked.

patients received gonadotropin-releasing hormone antagonist treatment for 3 cycles at 0, 4, and 8 weeks after surgery.

2.3. Statistical analyses

IBM SPSS Statistics 27.0.1 (International Business Machines Corporation, Armonk, NY) was used for data analysis. Variables with a normal distribution are shown as the mean ± standard deviation. Variables without a normal distribution are presented as medians (range).

3. Results

The baseline clinical characteristics of the 4 patients are shown in Table 1. The average age at diagnosis was 14.5 ± 3.3 years. All of them had periodic abdominal pain for an average of 5.5 ± 2.1 months. Physical examination revealed a well-developed labium majus and labium minus, normally positioned urethral meatus, and vaginal atresia that accommodated only one fingertip. Ultrasound and magnetic resonance imaging examinations

Table 1
Clinical characteristics of patients with type II vaginal atresia.

Patient number	1	2	3	4
Age, yr	12	12	15	19
Duration of periodic abdominal pain, mo	8	3	5	6
Uterine malformation	None	Unicornuate uterus	None	None
Urinary malformation	None	None	None	None
Endometriosis	None	None	None	Yes

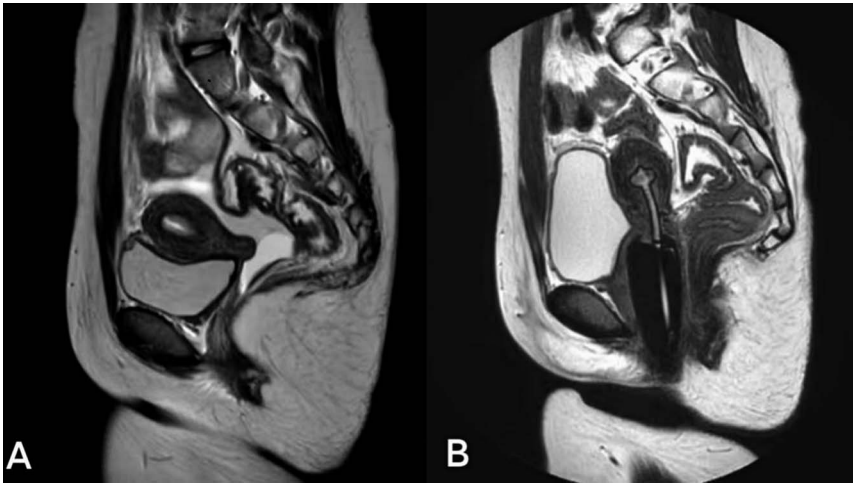


Figure 3. MRI of patients with type II vaginal atresia before and after the surgical procedure. (A) MRI of patient with type II vaginal atresia before the surgical procedure. (B) MRI of patient with type II vaginal atresia after the surgical procedure. MRI = magnetic resonance imaging.

revealed that all patients had type II vaginal atresia with adequate uterine corpus development, except for 1 patient who had a unicornuate uterus. Figure 3 shows magnetic resonance images of a patient with type II vaginal atresia before and after surgery. All patients had hematometra of different sizes in the pelvis, whereas 1 patient had endometriosis. No abnormalities were observed in the structure of the urinary system in any patient. The chromosomal examination results of all 4 patients were normal (46,XX). After the surgical procedure, all patients experienced regular menstruation. The average follow-up was 20.3 ± 8.7 months. The median vaginal lengths at the time of the surgical procedure and 6 months after the operation were 9.5 cm (range, 9.0–10.0 cm) and 7.0 cm (range, 7.0–7.5 cm), respectively. The average vaginal width at the time of the surgical procedure and 6 months after the operation were 3.08 ± 0.65 and 2.53 ± 0.39 cm, respectively. Until the end of follow-up, the length and width of the vagina coincided with those at 6 months postoperatively. No serious complications such as bleeding, infection, or urinary or rectal-anal injury occurred in any of the patients (Table 2). The vaginal mucosa had developed well, and the cervix and fornix had relatively normal shapes 4 months after surgery (Fig. 4). Six months postoperatively, patient 1 underwent vaginal dilation and adhesion separation because of 3- and 9-o'clock adhesions at the top of the vagina, and a suitable vaginal stent was used. Three to 6 months after the last

cycle of gonadotropin-releasing hormone antagonist treatment, all patients experienced regular menstruation, and only 1 patient experienced slight pain.

4. Discussion

Congenital vaginal atresia is an obstructive deformity of the reproductive tract, and a functional endometrium often leads to endometriosis. Cases of vaginal atresia are rare, with an incidence of

Table 2				
Outcomes of surgical procedure in patients with type II vaginal atresia.				
Patient number	1	2	3	4
Complications				
Bleeding during surgical procedure (>500 mL)	None	None	None	None
Infection	None	None	None	None
Urinary injury	None	None	None	None
Anal and rectum injury	None	None	None	None
Tissue adhesion at top of the vagina	Yes	None	None	None
Vagina length at surgical procedure, cm	9.0	10.0	10.0	9.0
Vagina length 6 mo of postoperation, cm	7.0	7.0	7.5	7.0
Vagina width at surgical procedure, cm	4.0	3.0	2.8	2.5
Vagina width 6 mo of postoperation, cm	2.9	2.7	2.5	2.0
Duration of follow-up, mo	10	17	30	24

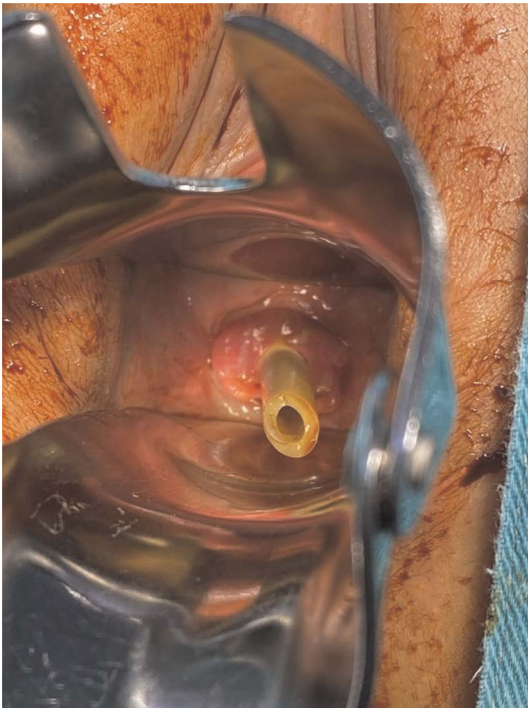


Figure 4. Physical examination of patients 4 months after surgery. The vaginal mucosa had developed well, and the cervix and fornix had relatively normal shapes 4 months after surgery.

0.001%–0.025%.^[6] There are 2 types of vaginal atresia. The incidence of type II vaginal atresia is rarer than that of type I. The longer the medical history, the more severe the symptoms of endometriosis. The essential principle of treatment is that once diagnosed, the procedure should be performed as soon as possible.^[7] Although patients with type II vaginal atresia can have a satisfactory sexual life after vaginoplasty and hysterectomy, the demand for fertility preservation is increasing in patients with better-developed uteri. In the past, assisted reproductive technology was immature, and pregnancy and delivery rates were low in patients who underwent uterine-preserving surgery. With the development of assisted reproductive technologies, uterine-preserving surgery has become increasingly critical for future pregnancies.

Studies have demonstrated that fertility preservation complicates surgery in patients with type II vaginal atresia,^[8] and only a few successful cases of fertility preservation among patients with this disease have been reported.^[9] The existing surgical approach involves creating a cavity in the vaginal area, that is, separating the cavity between the urethra and rectum to form an artificial cavity and avoid damage to the urethra, bladder, and rectum during the operation. With increasing demand for fertility preservation, surgical procedures for uterine preservation in these patients are currently in development. The challenge faced during uterine-preserving surgical procedures is the opening of the channel between the cervix and uterine cavity. In previous studies,^[5,10–17] there have been several ways to connect the uterine cavity with the neovagina (Table 3). Laparoscopy assists in the penetration of the artificial vaginal and uterine cavities, but it often requires incision of the fundus of the uterus to determine the position of the blind end of the lower uterine cavity and the orientation of the vaginal penetration and incision. Alternatively, the cervix and part of the anterior uterine wall are incised through the vagina to expose the uterine cavity and then sutured after the placement of a drainage tube. This undermines the integrity of the uterine corpus and cervix-centered ring to some extent.

In this study, we successfully connected the uterine cavity and neovagina while maintaining the integrity of the uterus and cervix under laparoscopic and transvaginal ultrasound monitoring. The median vaginal length and width at the time of the surgical procedure and at 6 months or even 3 years postoperatively were satisfactory. Small intestine submucosal biological patches are mainly made of porcine small intestine submucosa and can be used for host cell proliferation, tissue remodeling, and vascular regeneration as well as to support and enhance tissue repair.^[18] No serious complications, such as bleeding, infection, or urinary or rectal-anal injury, occurred in any patient, and all patients experienced regular menstruation soon after surgery. According to previous studies, granulomatous polyps, infections, and stenosis were the primary complications.^[10,11,14,17] Owing to the toughness of the atretic cervical tissue and the lack of protection of the columnar epithelium after cervical incision, postoperative cervical adhesion and atresia are very likely to occur and require reoperation; even the uterus could not be preserved in this case. Deffarges et al.^[19] reported 1 case of secondary stenosis of the anastomosis among 18 patients. Selvaggi et al.^[20] reported 1 case of stenosis at the anastomosis site and 1 case of granulomatous polyps in 2 patients. Ghafarnejad et al.^[13] reported 3 cases of stenosis and 2 cases of infection among 6 patients. Kriplani et al.^[11] reported 1 case of genital infection and restenosis, whereas hysterectomy was performed in 14 patients. In studies using sigmoid vaginoplasty, anastomotic leakage was also an unusual complication, with an occurrence rate of 0%–7.1%.^[17,21]

In our method, the cervical canal is directly punctured and penetrated by the puncture set for percutaneous nephrostomy with the assistance of laparoscopy. Laparoscopic surgery has

Table 3
Previous studies reporting the connection between uterus cavity and neovagina.

Author	Year	Number of cases	Approach to connect the uterus cavity and neovagina	Laparoscopic assistance	Integrity of uterus	Integrity of cervix	Complications	Pregnancy
Li et al. ^[14]	2013	2	The lowest pole of the uterine corpus was incised transversely.	Yes	no	Yes	Granulomatous polyps	-
Ghafarnejad et al. ^[13]	2013	7	Transverse incision was made in the most inferior part of the uterus or at a lower segment of the dilated cervix.	Yes	Only the integrity of the uterus or cervix can be preserved.	Yes	Neovaginal infection	One patient got pregnant and delivered at 37 wk.
Wu et al. ^[6]	2022	1	A puncture needle was used to puncture the cervix into the uterine cavity.	Yes	Yes	Yes	None	-
Abali et al. ^[12]	2013	1	An incision of 1.5-cm diameter was made on the uterine fundus, and the atretic cervical tissue was resected.	No	No	No	None	-
Kannayan et al. ^[10]	2009	7	Sigmoid colon conduit anastomosed to the posterior uterine wall after circular myomectomy.	No	No	No	Stenosis of the perineal neovaginal orifice	-
Alborzi et al. ^[17]	2023	7	An elliptical incision was made in the distal posterior lower segment of the uterine myometrium.	Yes	No	Yes	Proximal stenosis and vaginal discharge	-
Ding et al. ^[15]	2014	8	Direct incision was made on the atretic tissue until the uterine cavity was reached.	Yes	Yes	Yes	none	-
Kriplani et al. ^[11]	2012	9	A 0.5-cm breach was made in the uterine fundus using a monopolar needle.	Yes	No	Yes	Vaginal stenosis and purulent discharge	3 patients got pregnant and 1 delivered successfully
Fujino et al. ^[16]	2020	1	Fundal probe at the caudal end of the uterus is visible after incising through vaginal end. Skin biopsy punch of 3 mm was used to hollow out and resect the external cervical os; communication between the cervical canal and dilated uterine cavity was created.	Yes	Yes	No	None	-

several advantages. First, it can help avoid damage to lateral organs, such as the bladder and rectum. Second, it can assist in the diagnosis and treatment of coexisting pelvic diseases, such as endometriosis. Finally, it results in rapid recovery and a short hospital stay.

In our innovative approach, it is equally crucial that the puncture is guided by transvaginal ultrasound under laparoscopic monitoring. The puncture needle accurately reached the uterine cavity through the atretic cervix and gradually expanded through the cervical dilator. As an artificial vaginal cavity has formed, the anterior or posterior fornix can be opened laparoscopically to pull the cervix downward. It is more convenient for surgeons to expand the cervical canal further through the uterine dilator under direct vision.

Several studies have reported on the subsequent pregnancies after surgery. In a study of 42 patients,^[22] 11 attempted to become pregnant, 9 became pregnant, and 2 achieved successful delivery. Deffarges et al.^[19] reported that 4 of 18 patients had 6 spontaneous pregnancies. Ghafarnejad et al.^[13] reported that 1 of 7 patients became pregnant and delivered at 37 weeks. Karthik et al.^[23] reported that 1 patient became pregnant and delivered successfully. In our study, the follow-up time was limited, and no patient attempted to conceive. In the future, we intend to follow-up with these patients on a regular basis and monitor their pregnancy developments.

This innovative approach, laparoscopic and ultrasound-guided cervical canal penetration after vaginal reconstruction, is easy and feasible, preserves the integrity of the uterine corpus and cervix, and causes less harm to patients. However, this study also has limitations. The sample size of only 4 patients is small, hampering a comprehensive understanding of the surgical approach and an accurate assessment of its long-term effects. Future research should expand the sample size and extend the follow-up period to fully evaluate the method's effectiveness and safety. Nonetheless, we anticipate this novel technique will be verified in more clinical research centers, potentially benefiting patients with type II vaginal atresia more substantially.

5. Conclusions

Laparoscopic and ultrasonography-guided cervical canal penetration after vaginal reconstruction is feasible in patients with type II vaginal atresia and adequate uterine corpus development. There are a few side effects, but there are no obvious sequelae. Further studies are required to verify the long-term impact on quality of life and fertility in these patients.

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None.

Statement of ethics

This study was approved by the Ethics Committee of our hospital, with an approval number 2022K006. The patients and guardians were fully informed of the purpose of the surgical procedure and the possible complications before providing written consent to undergo the surgical procedure. All procedures performed in the study involving human participants were in accordance with the ethical standards of the institutional and national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Conflict of interest statement

The authors declare that they have no conflicts of interests.

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Author contributions

LX: Participated in data collection, writing of the manuscript, and data analysis;

LZ, CS, and XY: Participated in data collection and proofreading; BL: Participated in performance of the surgery, design of the study, draft revision, and final approval of manuscript to be submitted;

LL: Participated in assistance to the performance of the surgery;

XJ: Participated in performance of the surgery;

LC: Participated in assistance to the performance of the surgery.

Data availability

Data sharing not applicable to this article as no datasets were generated or analyzed during the current study.

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