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Usefulness of Endoscopy for Female Genital Plastic Surgery: A Series of 4 Cases with Genital Malformations

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

BEF Keiyo Imaeda
ABEF Yusuke Kobayashi
E Kanako Nakamura
B Shigenori Hayashi
B Tohru Morisada
E Kouji Banno
E Daisuke Aoki

Department of Obstetrics and Gynecology, Keio University School of Medicine, Tokyo, Japan

Corresponding Author: Yusuke Kobayashi, e-mail: kobax@a2.keio.jp
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Conflict of interest: None declared

Case series
Patients: Female, 18-year-old • Female, 25-year-old • Female, 12-year-old • Female, 29-year-old
Final Diagnosis: Mayer-Rokitansky-Kuster-Hauser syndrome • OHVIRA syndrome • Wunderlich syndrome
Symptoms: Excessive menstruation • irregular menstruation • lower abdominal pain • primary amenorrhea
Medication: —
Clinical Procedure: —
Specialty: Obstetrics and Gynecology

Objective: Congenital defects/diseases

Background: Female genital plastic surgery is rare and requires specific patient anatomical information to successfully carry out the appropriate procedure for each female genital malformation. Performing endoscopic surgery can increase the amount of information intraoperatively. We report on our experience of proactively using endoscopy to obtain additional intraoperative information to perform the appropriate surgical procedure suited to each female genital malformation, and evaluate the usefulness of concurrent endoscopy for improved surgical outcomes.

Case Reports: In Case 1, an 18-year-old woman underwent colpoplasty for Mayer-Rokitansky-Küster-Hauser syndrome using the laparoscopic Davydov procedure, wherein the pelvic peritoneum was precisely dissected using light from a laparoscope as a guide. In Case 2, a 25-year-old woman presented with suspected Wunderlich syndrome. Since the junction of the affected uterus with the normal uterus was unclear on preoperative imaging, we performed total hysterectomy of the affected uterus after identifying the anatomical structure by hysteroscopy. In Case 3, a 12-year-old girl with obstructed hemivagina and ipsilateral renal anomaly (OHVIRA) syndrome underwent transvaginal fenestration of the obstructed vaginal wall to prevent future menstrual molimen. Simultaneous hysteroscopy and laparoscopy allowed us to determine the location of a unilateral vaginal wall cyst and drain it safely. In Case 4, a 29-year-old woman had OHVIRA syndrome, and we conducted a laparoscope-based fenestration of the cervical canal safely and accurately.

Conclusions: Our series showed how, even for gynecological malformations without established standards for surgical procedure, a safe and minimally invasive surgery can be ensured if the amount of anatomical information is increased via endoscopy.

Keywords: Endoscopy • Genitalia, Female • Mullerian Ducts • Solitary Kidney

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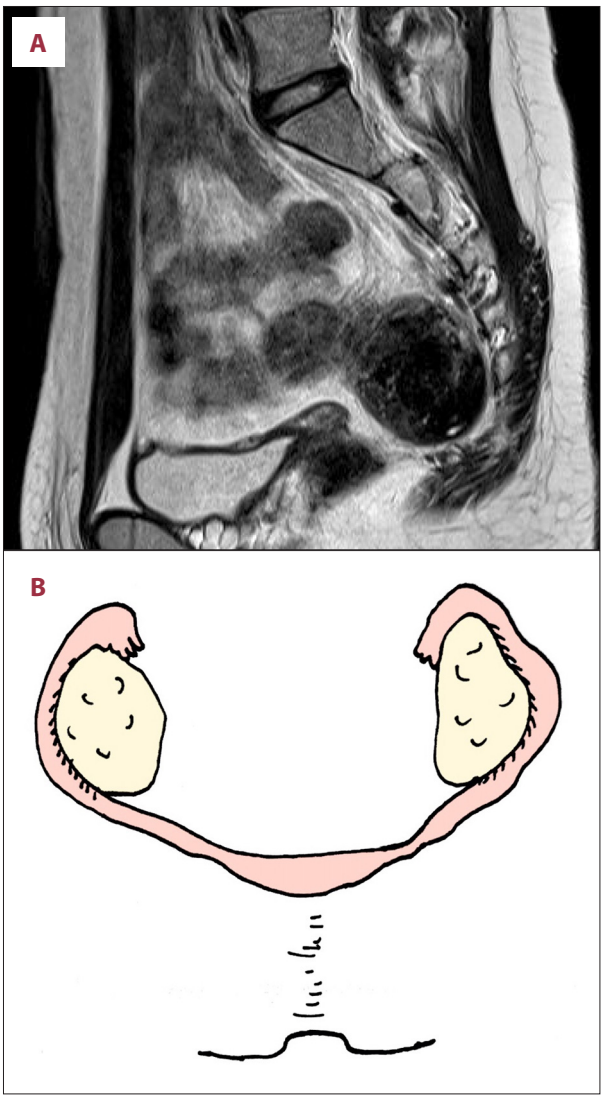
Background

Malformations of the female genital organs in adolescence and the reproductive years require surgery in some cases to improve functioning and quality of life. Due to the rarity of such cases, there are no established standardized surgeries [1]. Therefore, it is difficult to develop typical surgical approaches and conduct preoperative evaluation. At our hospital, we proactively use endoscopy to obtain additional intraoperative information to perform the appropriate surgical procedure suited to each individual patient. We retrospectively evaluated the usefulness of concurrent endoscopy for improved surgical outcomes in 4 patients who had undergone surgery for female genital malformations at our hospital.



Case Reports

Case 1

Case 1 involved an 18-year-old woman whose main problem was primary amenorrhea. Medical and imaging examinations revealed vaginal aplasia with rudimentary uterus, and she was diagnosed with Mayer-Rokitansky-Küster-Hauser (MRKH) syndrome (Figure 1A, 1B). After dilating the vaginal orifice for 2 years using a Hegar dilator, surgery was performed.

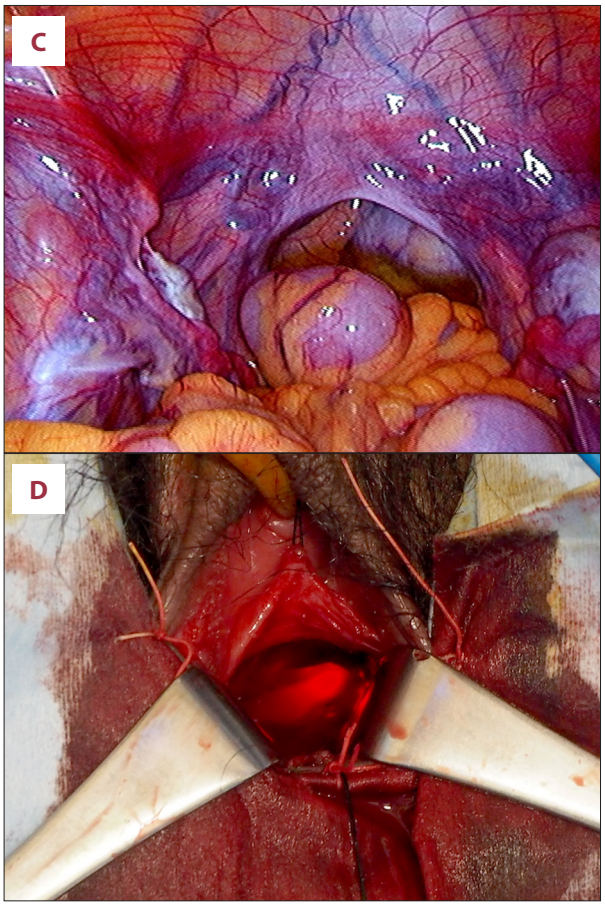


Figure 1. (**Case 1**). (**A**) MRI findings (T2-weighted imaging, sagittal plane) revealed vaginal aplasia and rudimentary uterus. Mayer-Rokitansky-Küster-Hauser syndrome was diagnosed. (**B**) Schematic representation of the genital organs seen in Case 1. (**C**) Intraoperative findings revealed that blood vessel damage could be avoided by advancing the scope to the blind end of the vagina between the bladder and rectum and then bluntly dissecting the connective tissue between the bladder and rectum in the direction of the light. (**D**) Use of the light of the laparoscope as a landmark for blunt dissection of the connective tissue between the bladder and rectum from the vaginal side.

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Surgical Procedure: Laparoscopy-Assisted Colpoplasty (Laparoscopic Davydov Procedure)

First, laparoscopy revealed a rudimentary uterus, and then the laparoscope was guided to the end of the peritoneum between the bladder and rectum (Figure 1C). Using the light from the laparoscope through the vagina as a landmark (Figure 1D), the vulvar vestibule was sectioned and the connective tissue between the bladder and rectum was bluntly dissected from the vaginal side. After reaching the pelvic peritoneum, a Cusco's speculum was inserted into the vagina and was opened in up-down and left-right motions to sufficiently dissect the surrounding tissue from the pelvic peritoneum. After opening the peritoneal sac, the peritoneum was pulled to the vaginal side and sutured to the vaginal opening. In this process, the blood vessels are clustered around the rudimentary uterus in patients with MRKH syndrome, so it is important to carefully evaluate the running of the vessels that are in close proximity (Figure 1C). Thereafter, a prosthesis was inserted into the newly created vagina, and the superior portion of the pelvic peritoneum was closed laparoscopically, thereby forming the blind end of the vagina. Prosthesis was used until the vaginal epithelium regenerated, and then a dilator was used to prevent vaginal restenosis.

Case 2

Case 2 involved a 25-year-old woman who presented with uterus didelphys and right renal agenesis, and suspected Wunderlich syndrome. The patient previously underwent resection of the vaginal wall and drainage for menstrual molimen, but her symptoms recurred. Magnetic resonance imaging (MRI) could not clearly identify the positional relationship of the uterus didelphys (Figure 2A); therefore, laparoscopic surgery was planned to assess the subject's anatomy precisely and to perform proper drainage.

Surgical Procedure: Laparoscopic Hysterectomy of the Affected Side

Laparoscopic observations ascertained that the right affected uterus was connected to the parauterine connective tissue of

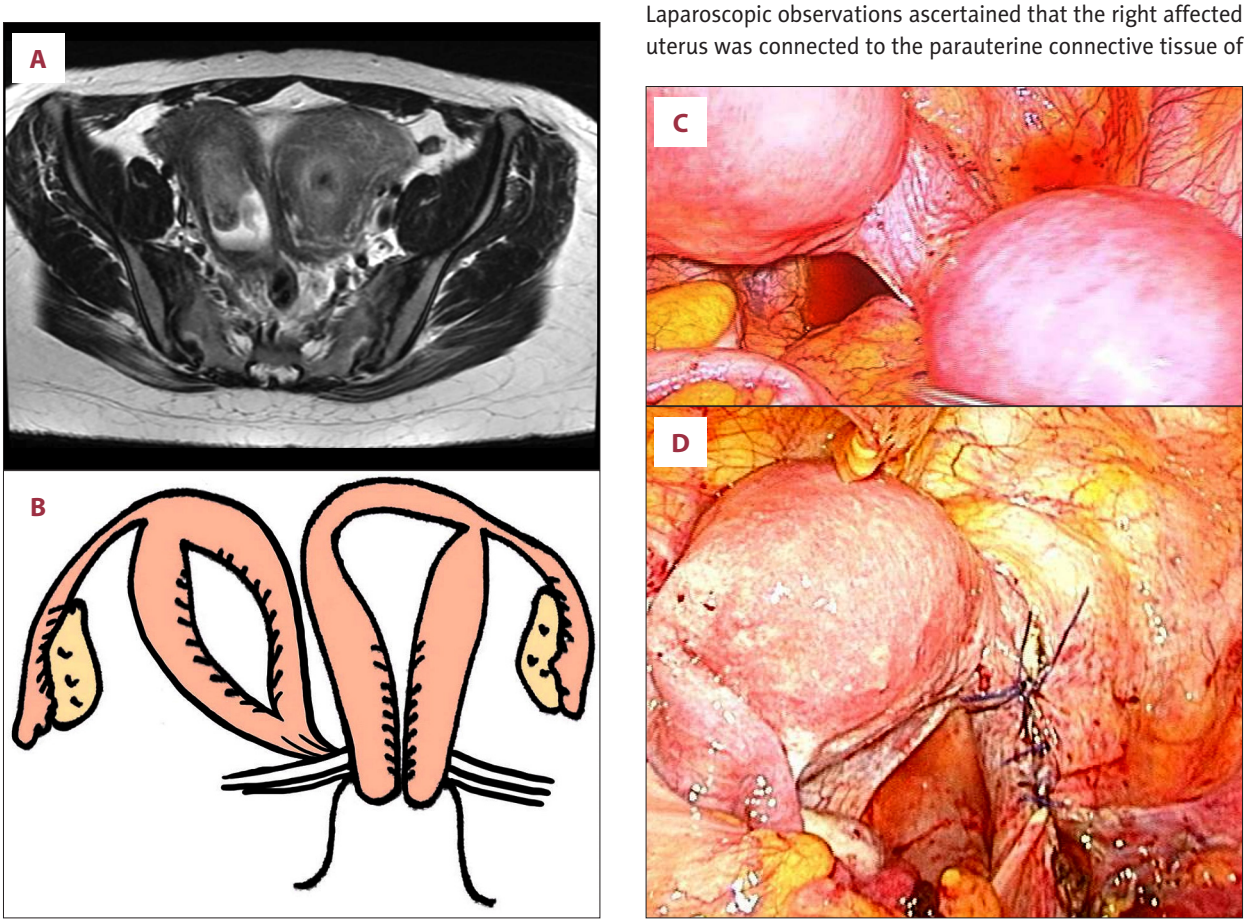

Figure 2. (**Case 2**). (**A**) MRI findings (T2-weighted imaging, axial plane) revealed duplicated uterus; however, continuity between the uterine cavity and vaginal cavity on the right side could not be confirmed. (**B**) Schematic representation of the genital organs seen in Case 2. (**C**) Intraoperative findings revealed that the right affected uterus was connected to the parauterine connective tissue of the left normal uterus by a cord-like structure. (**D**) The affected uterus was resected and removed laparoscopically.

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the left normal uterus by a cord-like structure (Figure 2B, 2C). The affected uterus was resected and removed laparoscopically (Figure 2D). The absence of the right urinary tract due to agenesis of the right kidney was confirmed during surgery, and the operation was able to be performed without concern about possible ureteral injury. After surgery, there has been no recurrence of menstrual molimen.

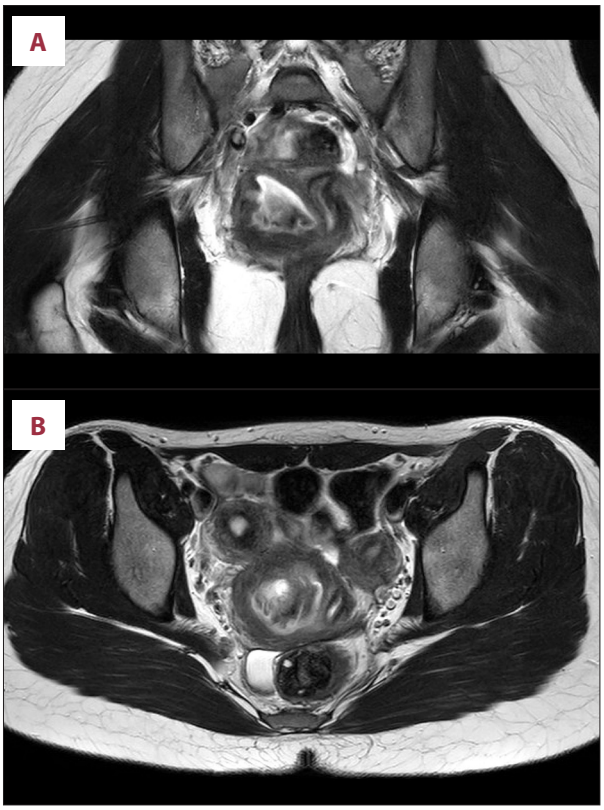
Case 3

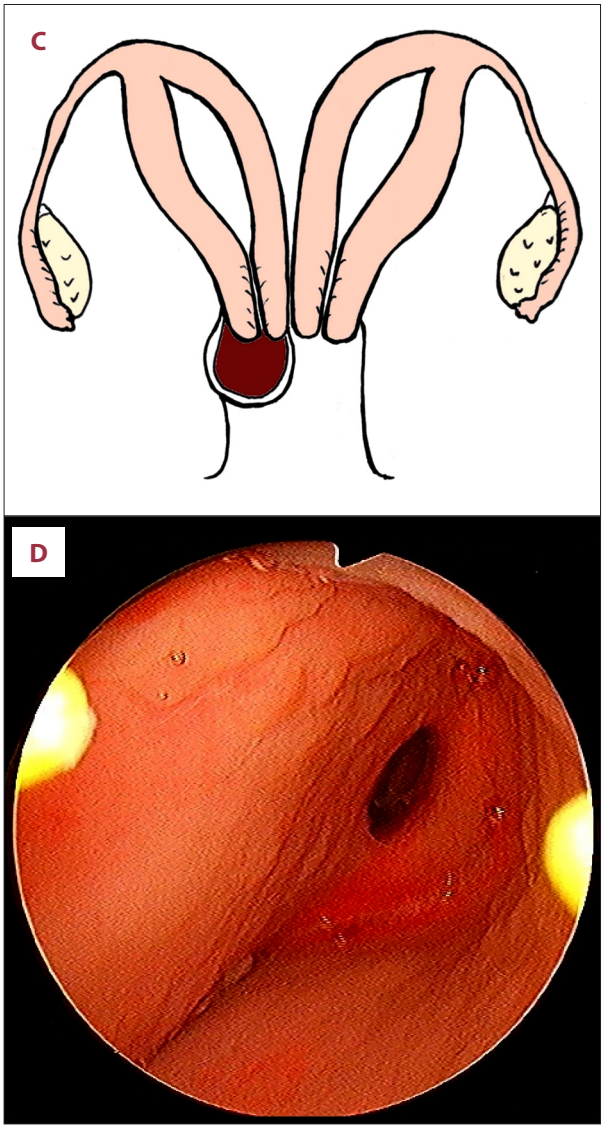
Case 3 involved a 12-year-old girl who reported excessive menstruation and lower abdominal pain. She presented with uterus didelphys, right vaginal closure, and right renal agenesis. The obstruction was so extreme that hemivagina and ipsilateral renal anomaly (OHVIRA) syndrome was suspected (Figure 3A-3C). We decided to use palliative surgery to reach a diagnosis and improve symptoms.

Surgical Procedure: Laparoscopy-Assisted Vaginal Cyst Fenestration + Hysteroscopy Examination

Since the patient was 12 years old, had no history of sexual intercourse, and presented a narrow hymenal ring, insertion of a hysteroscope into the vagina allowed for vaginal reflux fluid filling and distension of the vagina for intravaginal observation. Since hysteroscopy showed only the left os uteri, the right vaginal wall cyst was not visible (Figure 3D). We therefore decided to combine the hysteroscopy with laparoscopy. Laparoscopy confirmed the presence of uterus didelphys (Figure 3E) and, in combination with the hysteroscopic findings, the presence of a right vaginal wall cyst.

The combination of laparoscopy and hysteroscopy allowed us to perform fenestration drainage of the vaginal wall cyst (Figure 3F) while keeping track of the position of the vaginal wall cyst and the rectum. Then, we widened the hole which we had fenestrated and placed a Nelaton catheter (Figure 3G, 3H). A biopsy of the cyst's wall was obtained, and histopathological examination confirmed the presence of stratified squamous epithelium (Figure 3I), which allowed us to diagnose OHVIRA syndrome. Even 1 year after surgery, patency was maintained and no menstrual molimen was observed.





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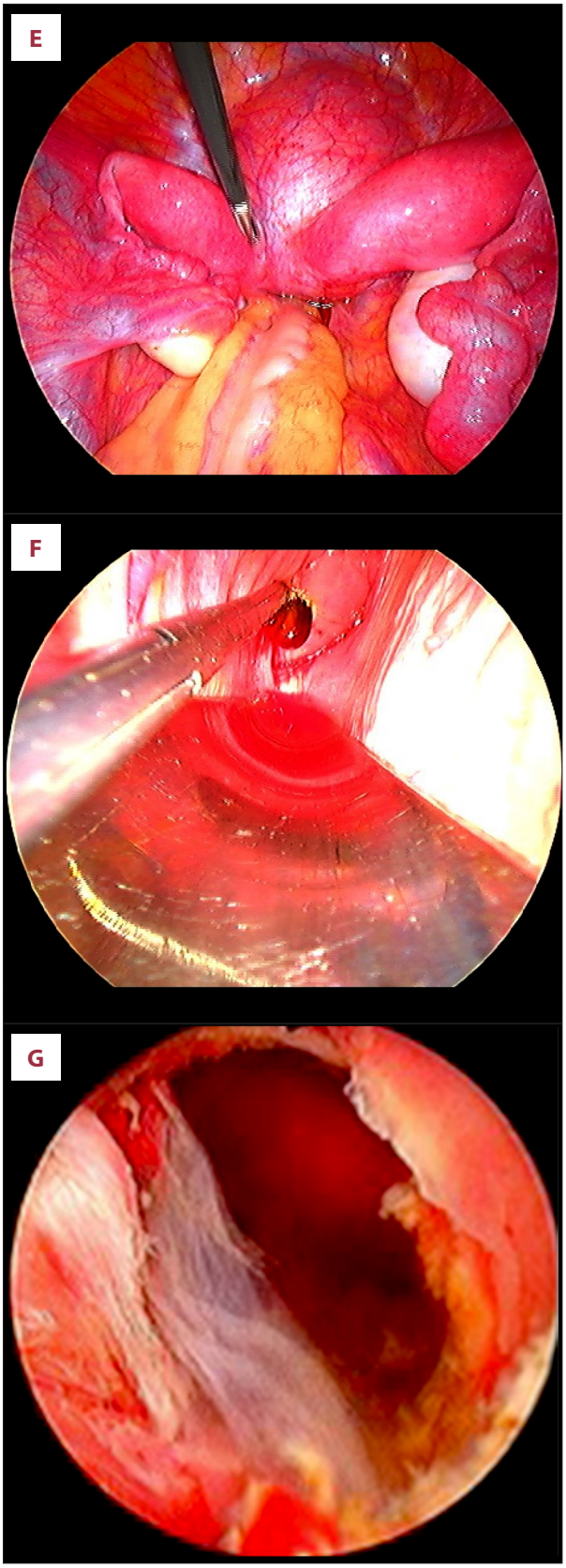

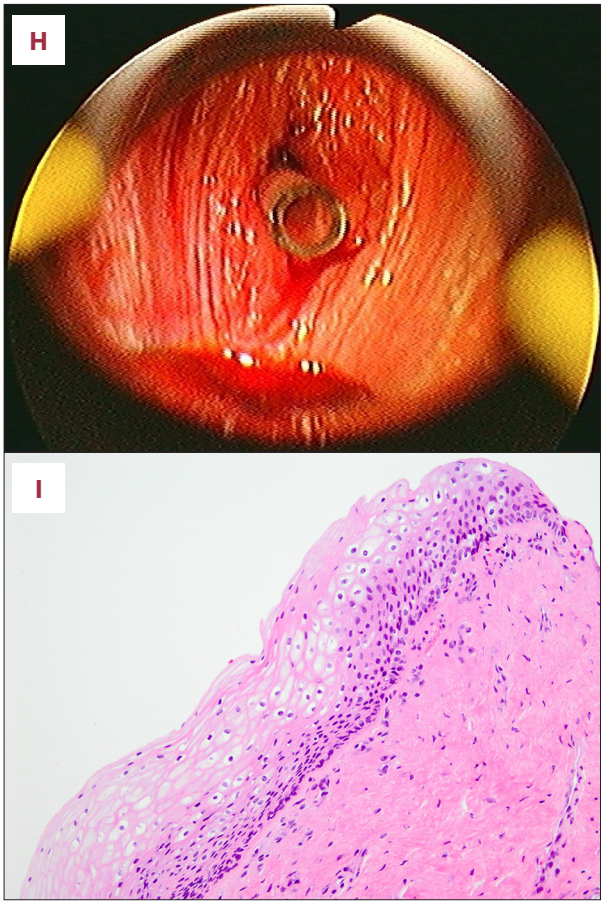

Figure 3. (**Case 3**). (**A, B**) MRI findings (T2-weighted imaging). Duplicated uterus and hemorrhagic cysts in the right vaginal wall, suspecting OHVIRA syndrome (**A:** coronal plane; **B:** axial plane). (**C**) Schematic representation of the genital organs seen in Case 3. (**D**) Only the left os uteri was seen. No holes in the right vaginal wall cyst could be observed. (**E**) Uterus didelphys and vaginal wall cysts were observed. (**F**) Simultaneous hysteroscopy and laparoscopy allowed us to perform puncture drainage of the vaginal wall cyst while keeping track of the position of the vaginal wall cyst and the rectum. (**G**) Depiction of the widening of the hole which we fenestrated. (**H**) A Nelaton catheter was inserted to prevent restenosis. (**I**) Histology of the cyst wall, composed of stratified squamous epithelium.

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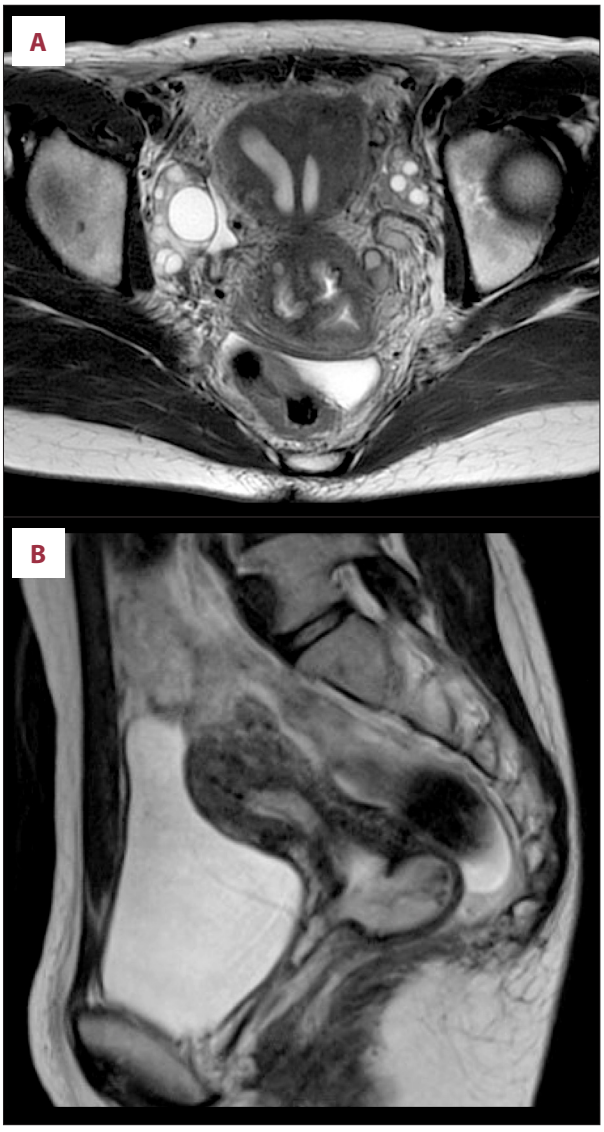
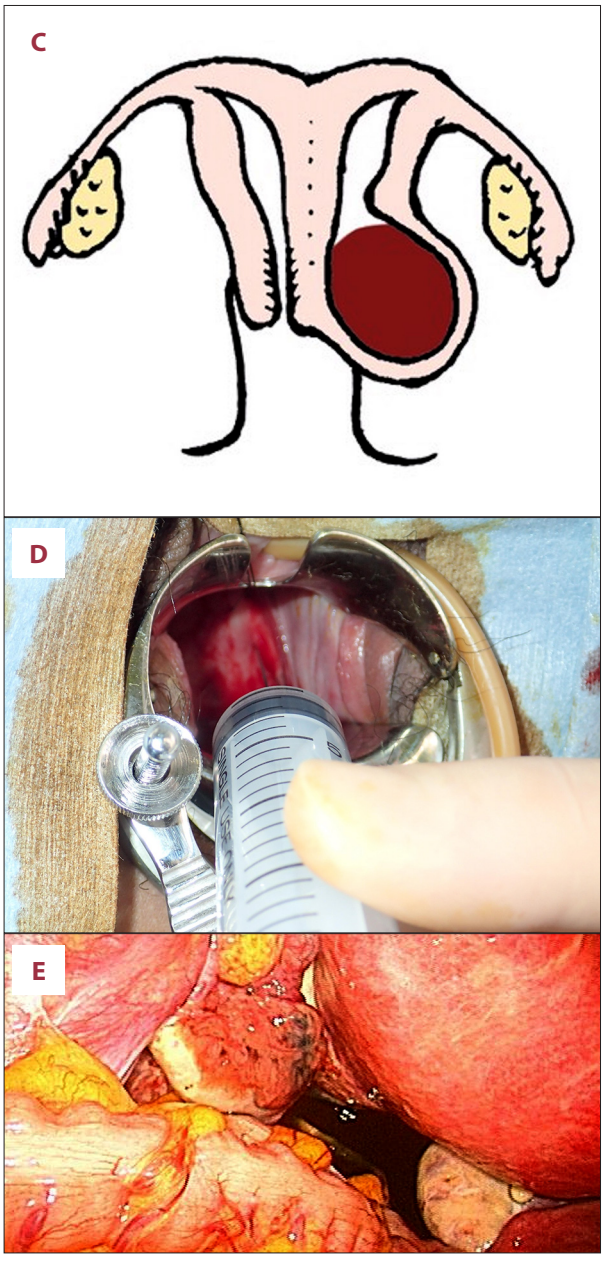
Case 4

Case 4 involved a 29-year-old woman who reported irregular menstruation and lower abdominal pain. MRI revealed a duplex uterus and a hemorrhagic cyst in the cervix of the left uterus (Figure 4A-4C). Furthermore, the patient had left renal aplasia and was suspected to have OHVIRA syndrome.

Surgical Procedure: Laparoscopy-Assisted Vaginal Cervical Duct Fenestration + Pigmented Fluid Flow Test

We performed an exploratory transvaginal puncture at the site where we expected to find the uterine cervical cyst (Figure 4D). After injecting indigo carmine, pigment outflow from the left tubal fimbriae was laparoscopically confirmed (Figure 4E), assuring us that we could puncture safely and insert a catheter into the cavity of the affected uterus. Such an approach eliminated the risk of damage to the surrounding tissue with a scalpel, and ensured that at the same site as the exploratory puncture the cervical opening and fenestration could be performed safely and accurately (Figure 4F). Furthermore, achieving sufficient fenestration allowed insertion of a Nelaton catheter (Figure 4G). Specifically, we used a "Robinson Nelaton catheter A® 13EG," which is relatively large, with an external diameter of 7.5 mm. The catheter was left in place for 1 month after the surgery, and the fenestrated vaginal wall has remained free of stenosis for 2 years after the surgery. The pathological examination of the fenestrated vaginal wall showed stratified squamous epithelium (Figure 4H), which led to the diagnosis of OHVIRA syndrome.





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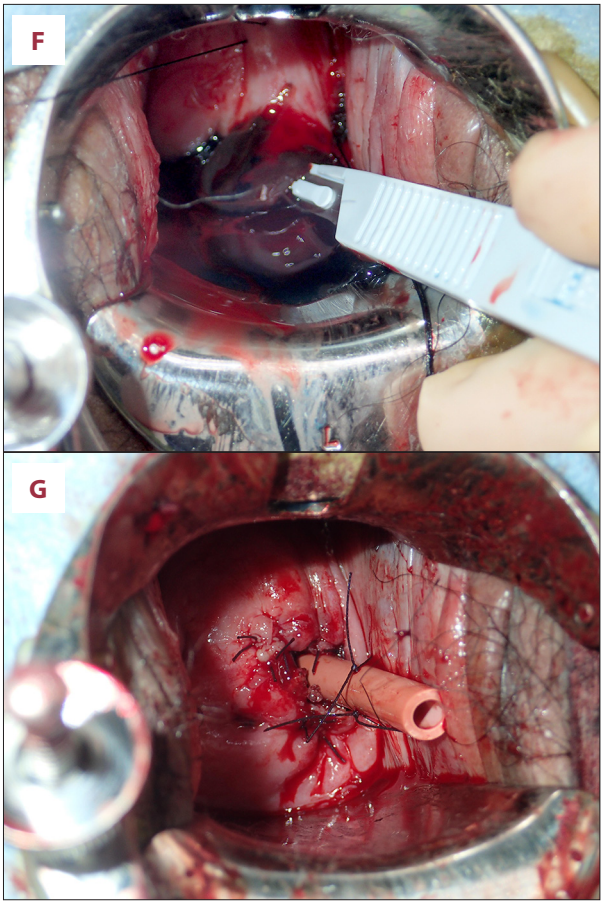

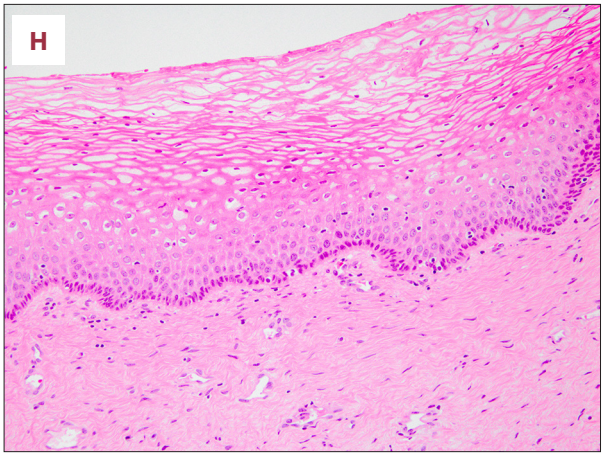

Figure 4. (**Case 4**). (**A**) MRI findings (T2-weighted imaging, axial plane): Duplicated uterus was observed. (**B**) MRI, sagittal plane: A hemorrhagic cyst was observed in the uterine cervix of the affected side. (**C**) Schematic representation of the genital organs seen in Case 4. (**D**) Exploratory puncture was performed at the site where the uterine cervical cyst was expected, and indigo carmine was injected. (**E**) Pigment outflow was laparoscopically observed from the left tubal fimbriae, thereby confirming that the puncture site had entered the uterine cavity of the affected side. (**F**) Spreading of the puncture site with a scalpel. (**G**) Insertion of a large Nelaton catheter to prevent restenosis. (**H**) Histology of the fenestrated vaginal wall, composed of stratified squamous epithelium.

Discussion

Female genital plastic surgery is very rare and there are no standard surgical techniques. Therefore, we performed endoscopy to gather additional information during surgery to ensure that the procedure was appropriate to each individual case. Here, we report our surgical precautions and emphasize the potential of endoscopic surgery to inspire surgery for various other female genital deformities.

In Case 1, some tips regarding the laparoscopic Davydov procedure for MRKH syndrome were demonstrated. MRKH syndrome is characterized by primary amenorrhea, congenital vaginal aplasia, and bilateral rudimentary uterus [2-4]. MRKH syndrome develops due to abnormal differentiation of the Mullerian ducts during the embryonic phase, but the precise cause remains unclear. The primary goal of treatment is to restore sexual function by performing colpoplasty. Various techniques are available [5-7], and new methods are constantly being devised. Using Frank's method, the mucosa of the vaginal vestibule is stretched using a device [6,8]. The most common method is the McIndoe procedure [9], wherein a skin graft from the abdomen, buttocks, or femoral region is used to create an artificial vagina. In the Ruge procedure [10,11], the sigmoid colon is used to create a vagina. In the Davydov procedure [12], the pelvic peritoneum is used. The Davydov procedure is minimally invasive and aesthetically pleasing, and enables sexual intercourse early after surgery. Furthermore, in recent years, colpoplasty combined with laparoscopic surgery performed to reduce the invasiveness associated with open surgery has gained widespread popularity, and primarily involves the laparoscopic Davydov procedure [12-15]. For these reasons, we selected the laparoscopic modified Davydov procedure for our patient. As mentioned above, the surgery was successfully performed owing to the creative use of the instruments in ways that differed from their original purpose, such as moving freely and directing the laparoscope's light to landmark the direction of peritoneal dissection, as well as the use of opening and closing movements of the Cusco's speculum to dissect the peritoneum.

In Case 2, the patient presented with Wunderlich syndrome. In 1971, reported cases of uterus didelphys presenting with communicating intrauterine cavity and renal agenesis on the same side as Gartner's duct cysts were termed Herlyn–Werner syndrome. In 1976, cases of duplex uterus accompanied by paracervical cysts (non-communicating unicornuate uterus) and ipsilateral renal agenesis were reported as Wunderlich

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syndrome [16-19]. At present, cases of duplex uterus, double vagina, obstructed hemivagina, ipsilateral renal agenesis, and cervical cysts are reported as Herlyn–Werner–Wunderlich syndrome (HWWS) [16,18,20,21]. Symptoms of HWWS often include lower abdominal pain gradually increasing after menarche and lower abdominal masses in adolescence [22]. Treatment for hematometrocolpos involves resection of the vaginal septum and transvaginal drainage of the blood. Recurrent hematometra caused by recurrent obstruction of the vaginal septum increases the risk of exacerbation of dysmenorrhea, and endometriosis can develop from the reverse flow of menstrual blood and endometrial cell transplantation (Sampson's transplant theory) [23,24], which is likely to lead to infertility. In Wunderlich syndrome, there have been many reported cases of restenosis and recurrence attributed to the fact that the cervical orifice is simply opened and formed [17]. Therefore, long-term regular follow-up observations and attention to repeated obstructions at the site of the vaginal wall fenestration are important [25]. In Case 2, we reported the process by which we were able to select the appropriate laparoscopic procedure for a patient with a subtype of anatomical anomaly similar to Wunderlich syndrome. In this case, a duplicated uterus and renal agenesis of the affected side with recurrent menstrual molimen were observed. Although Wunderlich syndrome was suspected, preoperative diagnosis was difficult because MRI did not show a clear anatomical position of the affected uterus with menstrual molimen. By performing laparoscopic surgery not only for treatment but also for diagnosis, we were able to clarify the precise anatomy of the affected uterus. In this case, we revealed that, unlike typical Wunderlich syndrome, the right affected uterus was connected with the parauterine connective tissue of the left normal uterus by a cord-like structure. Discovery of the precise anatomical structure by intraoperative laparoscopy was useful for determining the appropriate surgical procedure. The patient had repeated menstrual molimen, and she had 2 children, so we decided to perform a radical surgery.

OHVIRA syndrome, present in Cases 3 and 4, manifests with uterus didelphys, double vagina (accompanied by vaginal hematoma caused by obstruction on the affected side), and renal hypoplasia [25-27]. The syndrome is caused by impaired development of the Wolffian duct and non-fusion of the Mullerian ducts during the embryonic phase, with an incidence of 0.16-10% among malformations of the Mullerian ducts [28,29]. Clinical symptoms often include lower abdominal pain and dysmenorrhea [30,31], and treatment generally involves transvaginal fenestration of the obstructed vaginal wall to relieve symptoms and preserve fertility. The final diagnosis is confirmed by histopathological analysis of vaginal tissue obtained by biopsy during drainage of the hematoma [25]. When histopathological diagnosis reveals stratified squamous epithelium on both the obstructed and non-obstructed sides of the vaginal wall, OHVIRA syndrome is confirmed. In Case 3, the patient was young, and the first step was to perform a minimally invasive surgery for symptom relief and diagnosis. Concurrent hysteroscopy and laparoscopy allowed us to diagnose OHVIRA syndrome and perform a vaginal wall cyst fenestration to prevent future menstrual molimen. Preoperative images showed a duplicated uterus, right renal agenesis, and right vaginal wall cyst, and we considered the possibility of OHVIRA syndrome or Wunderlich syndrome. Initially, a hysteroscopy was performed, but the location of the vaginal wall cyst was unknown and puncture drainage could not be performed. Therefore, by combining hysteroscopy with laparoscopy, we were able to determine the anatomy of the vaginal wall cyst and its relation to the rectum and safely perform fenestration drainage. In Case 4, a duplicated uterus and left external orifice obstruction with a hemorrhagic cyst in the left cervical area were identified. Moreover, the left cervical duct needed to be opened for menstrual molimen. The blind vaginal procedure alone was considered unsafe because of the risk of injury to the surrounding tissue. Therefore, we decided to use laparoscopic assistance. In this case, exploratory puncture of the cervical cyst was performed in combination with laparoscopy, thereby enabling sufficient opening and fenestration of the cervix at the same site, as well as insertion of a large Nelaton catheter to prevent restenosis.

Conclusions

Female genital malformations are rare and they present with myriad variations; hence, there is no standardized surgery to address them. If surgical treatment is chosen, it is important to refer to the literature. However, each appropriate surgical procedure must be selected on a case-by-case basis, and preoperative imaging examinations are also useful. More anatomical information can be obtained intraoperatively by utilizing laparoscopy so that a surgical procedure fully tailored to each individual can be successfully achieved. While surgery for female genital malformations is rare, we believe that it can be performed safely in the future by performing laparoscopic surgery and confronting the important factors that require caution intraoperatively during surgery.

Declaration of Figures' Authenticity

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