

Neovagina in Mayer-Rokitansky-Küster-Hauser (MRKH) Syndrome: Vaginoplasty Using Ileal Flap

Neovagina na Síndrome Mayer-Rokitansky-Küster-Hauser (MRKH): Vaginoplastia Com Segmento Ileal Sem Pedículo

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ABSTRACT

Introduction: Surgical treatment of patients with malformations of the female genital tract is a complex problem and there are different techniques described in the literature. The goal of all these techniques is the reconstruction of a neovagina that is anatomically similar to a vagina, with adequate length to facilitate sexual functioning and with the lowest risk of possible complications. The aim of this study is to describe the surgical technique for the reconstruction of a neovagina from an ileal segment without a vascular pedicle.

Methods: Description of a surgical technique developed in our tertiary referral teaching hospital in a patient with Mayer-Rokitansky-Küster-Hauser syndrome.

Results: The vaginoplasty surgery using ileal flap was performed in three steps. In the first part of the intervention a laparoscopic hysterectomy with bilateral salpingectomy was performed. The second step consisted of isolating the ileal segment, ileal anastomosis and preparing the ileal segment. After the isolated segment was prepared, it was repositioned in a vagina mold to configure the neovagina. Finally, the third step included the adaptation of the vaginal mold with the ileal mucosa to the vesicorectal space.

Conclusion: Ileal vaginoplasty without a vascular pedicle is an option that can be used to reconstruct the vagina, because it provides an excellent tissue for vaginal replacement. This technique can be used in patients with genital malformations of the genital tract with absence or vaginal hypoplasia.

Keywords: 46, XX Disorders of Sex Development/surgery; Reconstructive Surgical Procedures; Vagina/surgery

RESUMO

Introdução: O tratamento cirúrgico de doentes com malformações do trato genital feminino é um problema complexo. Existem diferentes técnicas descritas na literatura cujo objetivo é a reconstrução de uma neovagina anatomicamente semelhante a uma vagina, com comprimento adequado para facilitar o funcionamento sexual e o menor risco de complicações possíveis. O objetivo deste estudo é descrever a técnica cirúrgica para reconstrução de uma neovagina a partir de um segmento ileal sem pedículo vascular.

Métodos: Apresentamos uma técnica cirúrgica desenvolvida num centro universitário terciário numa doente com síndrome de Mayer-Rokitansky-Küster-Hauser.

Resultados: A cirurgia de vaginoplastia com segmento ileal foi realizada em três etapas. Na primeira etapa de intervenção foi realizada uma histerectomia laparoscópica com salpingectomia bilateral. A segunda etapa consistiu no isolamento do segmento ileal, anastomose ileal e preparação do segmento ileal. De seguida, o segmento ileal isolado foi reposicionado num molde de vagina para configurar a neovagina. Finalmente, a terceira etapa incluiu a adaptação do molde vaginal com a mucosa ileal ao espaço vesico-rectal.

Conclusão: A vaginoplastia com segmento ileal sem pedículo vascular é uma opção que pode ser utilizada para reconstrução da vagina, pois proporciona um excelente tecido para reposição vaginal. Esta técnica pode ser utilizada em doentes com malformações do trato genital com ausência ou hipoplasia da vagina.

Palavras-chave: Procedimentos Cirúrgicos Reconstructivos; Transtornos 46, XX do Desenvolvimento Sexual/cirurgia; Vagina/cirurgia

INTRODUCTION

Vaginal reconstruction is important in some gynecological conditions with the vaginal agenesis syndrome, such as the Mayer-Rokitansky-Küster-Hauser syndrome (MRKH), insensitivity to androgens, and in transgender women.

Mayer-Rokitansky-Küster-Hauser syndrome is a rare disorder and has an incidence of 1 per 4500 to 5000 females.¹ It is caused by embryologic underdevelopment of the müllerian duct, with resultant agenesis or atresia of the vagina, uterus and/or fallopian tubes in the setting of a normal female karyotype.² Even though most cases appear to

be sporadic, there are some familial cases with an autosomal dominant mode of inheritance.³

Two types of MRKH syndrome have been described, referred to as type 1 and type 2. In MRKH type 1 syndrome only the organs of the female reproductive system are involved.⁴ Type 2 MRKH syndrome has additional congenital abnormalities which can include: kidney, skeletal, hearing and cardiac problems.⁵

Dysregulation of the *Wnt* and/or *Hox* genes may affect cell migration and differentiation of müllerian structures

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leading to the development of MRKH syndrome.⁶ The ovaries are typically normal in structure and function, due to their separate embryologic origin.

The MRKH syndrome is commonly diagnosed in adolescence or early adulthood and primary amenorrhea is often the first sign.⁷ In addition to infertility, another manifestation of Mullerian agenesis is difficulty with sexual intercourse due to the impossibility of vaginal penetration.⁷ Women with MRKH syndrome have a typical growth and normal pubertal development, presenting a karyotype 46,XX.⁸

Several treatment techniques have been employed to create a neovagina in these patients. A breakthrough procedure for vaginoplasty surgery was the Abbè-McIndoe technique, which uses a split-thickness skin graft to cover the neovaginal canal.⁹ Several other techniques have been proposed, such as Williams vaginoplasty (vulvovaginoplasty), Vecchietti (continuous traction applied through the abdominal wall in an 'olive' inserted into the vagina dimple) or Davydov procedure (peritoneal vaginoplasty). Another option is the use of intestinal segments, which was first described by Baldwin in 1904.¹⁰ Vaginoplasty using a sigmoid colon segment is another procedure performed by some pediatric surgeons and urologists.¹¹

The timing for the creation of a neovagina is elective, but treatment should be deferred until late adolescence when the patient is mature enough to agree to the procedure and to be able to adhere to postoperative dilation using different vaginal molds.¹² The treatment for these patients consists of creating a sexually functioning neovagina and supportive psychological care.

The aim of this study was to describe the surgical technique for the reconstruction of a neovagina.

METHODS

We present the surgical technique of vaginal reconstruction from an ileal segment without a vascular pedicle assisted by laparoscopy in a patient with MRKH syndrome and hypoplastic uterus. This technique was developed by a multidisciplinary team formed by gynecologists and general surgeons in our tertiary university center. Written informed consent was obtained from the patient for publication of this case report and the accompanying images.

RESULTS

Case report and surgery technique

A 22-year-old woman with MRKH syndrome presented with complaints of primary amenorrhea. She also reported that it was impossible to have normal vaginal intercourse with her male partner. The patient had depression and was on medication. Review of her pubertal development denoted an onset of thelarche (breast development) at 10 and pubarche (pubic hair development) at 11 years of age.

Her parents reported an unremarkable family history and denied congenital anomalies among family members. Her body mass index (BMI) was 22 kg/m². On gynecological examination, normal secondary sexual characteristics were observed; breast development was classified as Tanner stage 4. The examination revealed well developed labia and clitoris with a shallow vaginal pit and a short vaginal pouch, approximately 1 cm deep.

The patient's laboratory studies showed: blood karyotype 46XX, estradiol-17-beta 183.5 pg/mL (Follicular phase: 11.0 - 165.0; Ovulatory peak: 146.0 - 526.0; Luteal phase: 33.0 - 133.0; Postmenopause: 0.0 - 37.0), progesterone 21.37 ng/mL (Follicular phase: 0.15 - 1.40; Ovulatory peak: 3.34 - 25.56; Luteal phase: 4.44 - 28.03; Postmenopause 0.0 - 0.73), follicle-stimulating hormone 2.66 mIU/mL (Follicular phase: 2.5 - 10.2; Ovulatory peak: 3.4 - 33.4; Luteal phase: 1.5 - 9.1; Postmenopause 3.0 - 116.3), and luteinizing hormone 3.19 mIU/mL (Follicular phase: 1.9 - 12.5; Ovulatory peak: 8.7 - 76.3; Luteal phase: 0.5 - 16.9; Postmenopause 15.9 - 54.0).

A pelvic magnetic resonance imaging (MRI) revealed ovaries along with a rudimentary uterus and vagina expressed by a thin hyposignal line without an identifiable cervix. The rudimentary cavity had no hematometra (collection of blood in the uterus). The bladder, kidneys, urinary tract and rectum were normal in appearance.

Before the operative procedure, a thorough bowel preparation that included a one day low-residue semifluid diet, one day liquid diet plus one day fasting was performed. A cleaning enema was given on the day before surgery. Prophylactic antibiotic treatment (2 g of cephazolin) was administered intravenously two hours before the operation.

The vaginoplasty surgery using ileal flap was performed in three steps: (1) laparoscopic hysterectomy with bilateral salpingectomy; (2) isolation of the ileal segment, subsequent ileal anastomosis, preparing the ileal segment and adapting it to the mold; (3) creation of the neovagina, with

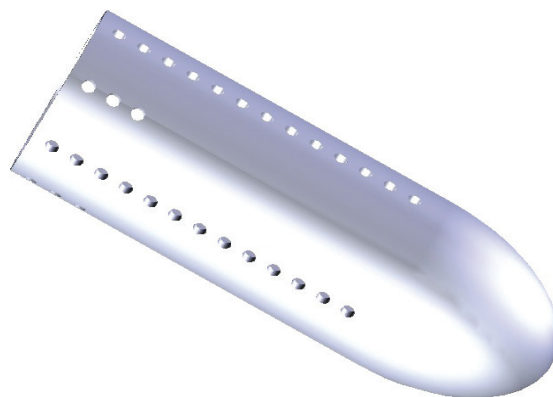


Figure 1 – 3D model of the vaginal mold

the adaptation of the vaginal mold with the ileal mucosa to the vesicorectal space.

A team of senior gynecologists and a general surgeon performed all procedures.

A previously designed vaginal mold in the form of a cylinder with a beveled end and a round one was developed (Fig. 1). It had eight rows of small holes to allow the tissue fixation sutures to pass through in order to suspend it to the walls of the neovagina.

We created different sizes of vaginal molds in order to adapt to the space that is dissected between the rectum and the bladder.

The patient was placed in the lithotomy position. Prior to the procedure, a Foley catheter was inserted in the bladder through the urethra to identify these structures and ensure they were well away from the dissection field. Laparoscopy was performed through four-ports: one 11-mm trocar was placed in the umbilicus and three 5-mm trocars were placed under laparoscopic visual guidance: one in each (left and right) iliac fossa and one in the midline, above the symphysis. The patient was then placed in the Trendelenburg position. The surgery was performed with the UltraCision® (UHS) harmonic scalpel (Smithfield, RI), as it is a safe and fast tool for tissue cutting and coagulation.

The surgery was performed according to the following steps:

Step I: Laparoscopic approach

1. For bilateral salpingectomy, the fimbriae were identified and grasped with an atraumatic forceps. The

fimbriated ends were suspended and elevated from the ovary. A bowel grasper was placed below the first one to prevent the lesion of the infundibular pelvic ligament. Electrocautery was used to detach the fimbria from the ovary and then continue across the mesosalpinx toward the uterus.

2. Tissue transection was continued until the adnexa were completely isolated from the uterine corpus. Next, the round ligament was coagulated and transected. Coagulation of the round ligament allowed opening the leaves of the broad ligament and entering the vesicouterine and retroperitoneal spaces and subsequent dissection. Dissection performed with UltraCision® in vascularized areas and Maryland dissector were used when separating planes. In this way, a hysterectomy was performed on the hypoplastic uterus (Fig. 2). It was necessary to increase the umbilical orifice up to 2 cm, in order to be able to exteriorize the uterus.

Step II: Preparation of the ileal segment and adaptation to the mold.

1. The team mobilized the small intestine intra-abdominally to obtain a free ileal loop. The mesentery was dissected from the ileal segment to an extent of 20 cm using bipolar energy when necessary.
2. This was removed from the abdominal cavity by laparoscopy through the umbilical orifice that was enlarged.
3. Outside the abdominal cavity, enterectomy was



Figure 2 – Laparoscopic image after bilateral salpingectomy and hysterectomy

performed and an ileal segment without a pedicle was isolated.

4. The continuity of the ileum was restored by a single layer of end-to-end anastomosis using GIA™ (Fig. 3).
5. Hemostasis was verified and if necessary, a suture or electrocautery was used to contain hemorrhage.
6. The removed bowel segment was prepared, with multiple washes using saline solution and isolated from the intestinal mucosa (removal of the parietal peritoneum). The segment was used as a single layer to form the neovaginal lining.
7. The prepared intestinal mucosa was adapted to the surface of the vaginal mold that was previously developed by the team (Fig. 4). The intestinal mucosa layer was in contact with the vaginal mold and the deperitonealized surface was facing outwards.
8. After adaptation to the mold, the vaginal mucosa at the end of the internal apex of the mold was sutured with 910 polyglactin thread.

Step III: Reconstruction of the neovagina

1. Bladder instillation of 300 cc of methylene blue and saline was performed to allow identification of bladder lesions in the following steps. To facilitate dissection, the vesicorectal space was infiltrated with

cold saline, adrenaline and lidocaine in the anterior wall of the vesicorectal space.

2. A longitudinal incision was made in the apex of the vaginal dimple.
3. The channel was dissected on each side of the median raphe. The space between the urethra and bladder anteriorly and the anus and rectum posteriorly was dissected, until a 3 cm wide and 7 cm long tunnel was obtained. The dissections were performed initially with monopolar energy and then blunt dissection was performed with Metzenbaum Scissors to minimize the risk of lesions in the bladder and rectum and preserve vascularization.
4. The procedure was controlled laparoscopically so that space was dissected until the lower surface of the peritoneum was reached. It is important to have a clear understanding of the anatomy and estimation of the space between the bladder and rectum and whether it will accommodate the neovagina, to avoid entering the peritoneal cavity and injuring adjacent organs.
5. We created different sizes of vaginal molds in order to adapt to the space between the rectum and the bladder that is dissected. The vaginal mold with the intestinal mucosa was irrigated with oxytetracycline

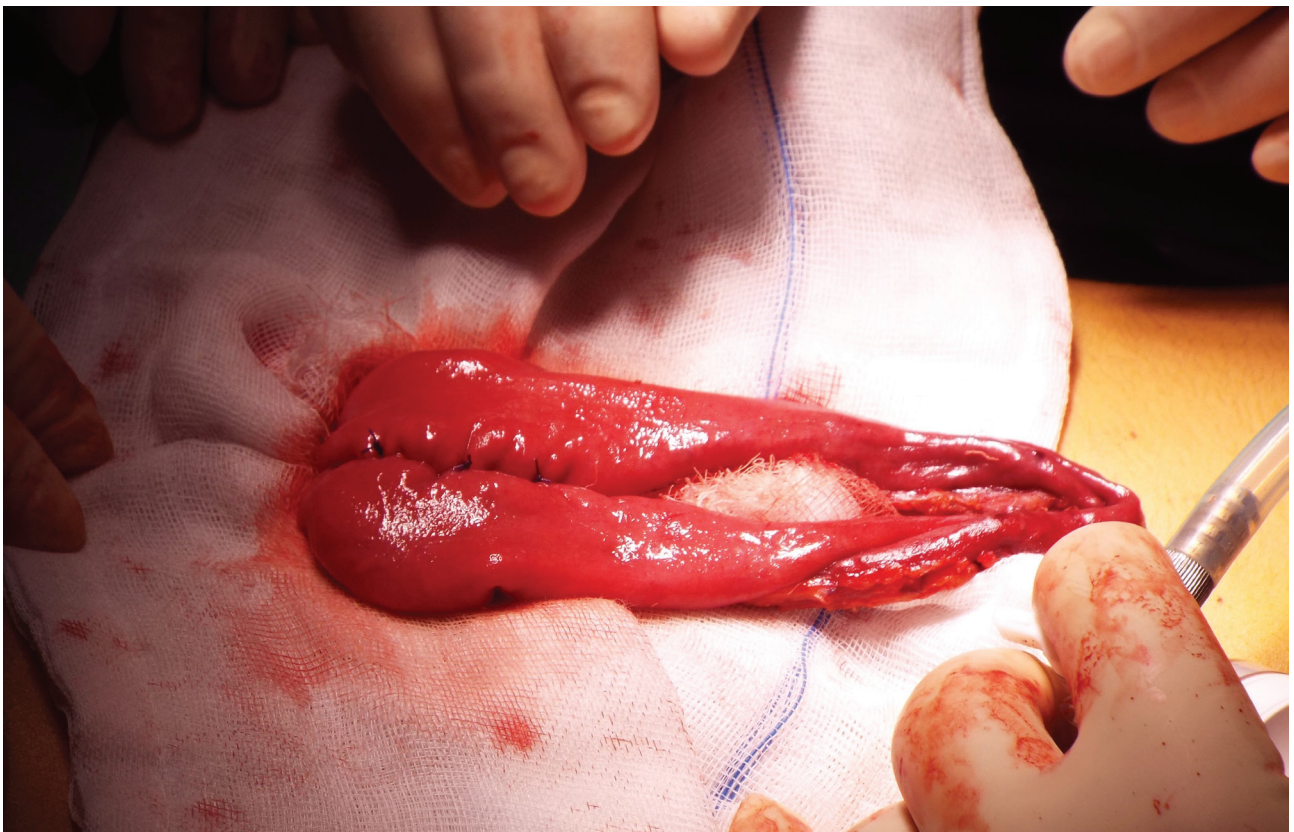


Figure 3 – Preparation for ileal anastomosis and application of GIA™

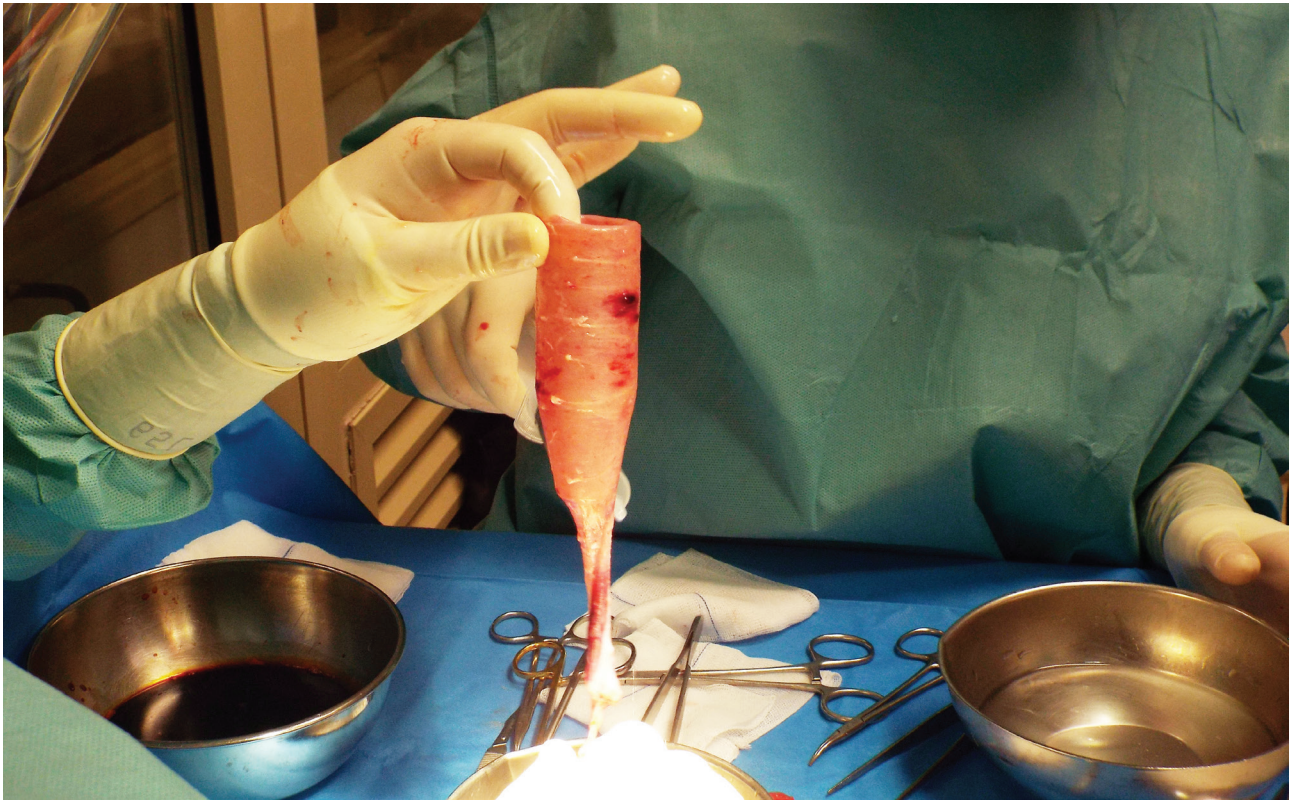


Figure 4 – The prepared intestinal mucosa was adapted to the surface of the vaginal mold

hydrochloride solution. Introduction of the vaginal mold and fixation to the labia majora was made with 2-0 sutures.

After surgery

1. Second-generation cephalosporin 1g 8/8 hours was given intravenously for two days.
2. The Foley catheter was removed in the third postoperative day, to minimize the risk of urinary retention.
3. After one week, the vaginal mold was removed under general anesthesia (Fig. 5).
4. At discharge, 10 days after surgery, the patient was instructed to wear the dilators for the whole night during the first three months. The creation of the neovagina is maintained using progressively larger dilators by increasing length and width as tolerated until a suitable vaginal dimension is established. The application of an estrogen based vaginal cream daily was used to promote the growth of the intestinal mucosa for three months.
5. The frequency of using vaginal dilators will be lower over time in patients with an active sex life.

After three months of treatment, the patient developed a 6 cm-long and 1.5 cm-wide diameter neovagina. The frequency of use of vaginal dilators was reduced at the end of the fourth month after the operation.

The patient was able to have comfortable sexual intercourse after the treatment, and she was satisfied with the results one year after the operation.

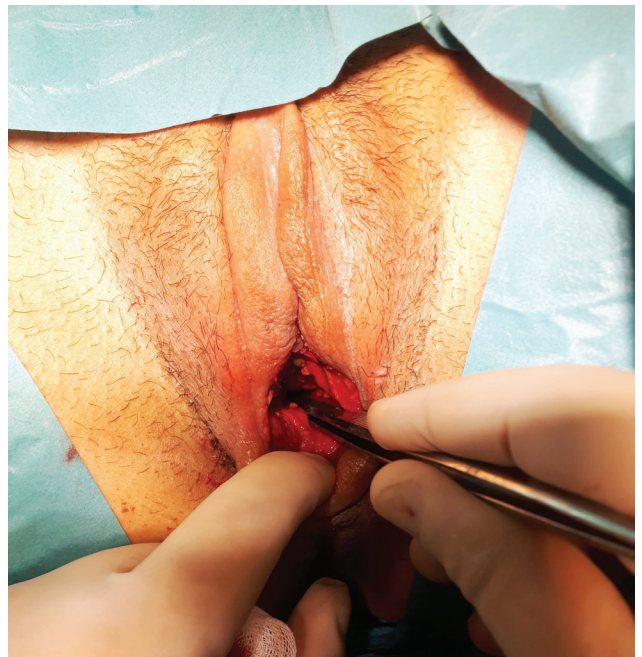


Figure 5 – Result after removal of vaginal mold, 1 week after surgery

DISCUSSION

The psychological effect of the diagnosis of müllerian agenesis should not be underestimated. There are three fundamental aspects with great impact on mental health, to be considered when evaluating these patients: amenorrhea, interference in sexual life and infertility.

Amenorrhea is a factor of anxiety, especially in adolescence when patients realize that they will not menstruate in a similar way as other women and that compromises fertility. Options for having children should be addressed with these patients, including adoption and assisted reproduction techniques with the use of gestational surrogacy. As the ovaries are present, patients with this condition can have biological children through in vitro fertilization. Uterine transplantation may be another option, but it is currently considered experimental and is not widely available.¹³

To establish a new and positive self-esteem, it is also necessary to construct a good functional vaginal substitute, which needs to be aesthetically pleasing, require low post-operative manipulation and provide long-term sexual satisfaction.¹⁴

Vaginoplasty is an important surgical procedure to improve physical and psychological well-being of women with congenital vaginal agenesis. MRKH syndrome may be diagnosed during adolescence, but treatment is usually delayed until the patient is ready to start sexual activity. We consider that there may be a greater motivation and consequently a better adherence to the recommendations in the postoperative period when the patient is sexually active, which contributes to the success of the treatment. Counseling and psychological preparation before treatment is also essential for its success.

Many methods of vaginal reconstruction have been reported in the literature. According to the American College of Obstetricians and Gynecologists, the progressive dilatation technique, described by Frank, should be attempted first before surgical intervention.¹⁵ Although satisfactory vaginal length and sexual function can be achieved, this method requires a long and persistent collaboration by the patient, and they may feel embarrassment and shame.¹⁶

Surgery is a second line treatment, usually reserved for patients who have failed the conservative approach or prefer surgery after a thorough discussion of the advantages and disadvantages of the different techniques.

The best surgical treatment of vaginal agenesis is still under discussion. Ideally, the reconstruction should be able to provide sufficient dimension, physiological mucosa lining, and satisfactory sexual function, while causing minimal donor site morbidity.

Techniques using the ileum and the cecum are described in the literature but are less performed when compared to the sigmoid colon.¹⁷ Unlike other cases described

in literature, in the case presented, a pedicle-free ileum segment was used to create the neovagina's inner lining. It was possible to obtain a neovagina with a length of 6 cm, after three months. Once a vaginal length of approximately 8 cm was achieved at six months after surgery, the patient began to have a normal sex life without experiencing discomfort during penile penetration.¹⁸

According to the literature, ileal segment vaginoplasty provides a cosmetically acceptable neovagina with a good width and depth without the long-term use of vaginal dilators.¹⁹ However, the question regarding the need for regular vaginal dilation after ileal vaginoplasty is debatable. We believe that it is not necessary to continue with routine vaginal dilation if the patients are sexually active.

Other factors associated with satisfactory results from different surgical techniques include natural lubrication facilitating sexual intercourse which decreases the incidence of dyspareunia, texture and appearance that is similar to that of a natural vagina, little tendency to shrink, the relatively stronger resistance to trauma, and reduced secondary deformity in the perineum.²⁰⁻²²

There are several reasons for using ileum instead of sigmoid colon for vaginal reconstruction. Not only does the ileum produce less mucus and a less intense smell than the large bowel²³ but it also has lower rates of potential chronic complications, for example, ulcerative colitis, hereditary polyposis²⁴ or primary adenocarcinoma,²⁵ as compared with the sigmoid segments.

It is also important to consider that the ileum is technically less demanding for use in laparoscopic assisted vaginal reconstruction because it has greater mobility, and the segment is easy to cut and anastomose. In addition, the growth of the ileum allows for surgery at an earlier age if indicated.²⁶

In most of the surgeries described in the literature, the resection of an ileal segment has a vascular pedicle.

Due to its mobility and constant vascularization, the ileal segment represents an option which is also valid for vaginal reconstruction. However, what we verified with our technique is that it is not necessary to isolate a pedicle in the flap used for the reconstruction of the vagina.

The small intestinal submucosa is formed by structural and functional proteins, such as collagen, fibronectin, glycosaminoglycans and growth factors.²⁷ This extracellular matrix favors good biological tolerance, with no tendency towards rejection.²⁸ It has low rates of infection and, according to some authors, even certain antibacterial properties.²⁹ It also induces angiogenesis and the prominent growth of cells of several strains.³⁰ The epithelialization observed in almost all the intestinal submucosa graft shows the good capacity of reepithelization of the intestinal submucosa.

The abundant neovascularization that accompanies

these situations contributes to the acceleration of the neovagina reconstitution process. The presence of angiogenic growth factors was evidenced, such as endothelial growth factor (EGF) and growth factor for fibroblasts (FGF).³¹

The fact that the patient is young and has no associated risk factors such as diabetes mellitus, smoking, hypertension or other cardiovascular disease favors successful neovascularization and consequently, flap viability.

The approach chosen was laparoscopy. The advantages of the laparoscopic technique when compared with laparotomy are the smaller abdominal incisions, no exposure of internal organs, reduced loss of blood and body fluids, quicker recovery of bowel function and lower rates of complications. Moreover, it allows a better visualization of the pelvic cavity and the innermost portion of the rectal-neovaginal space to avoid rectal lesions and reduce postoperative morbidity.²¹ Despite this, we performed the ileal anastomosis outside of the abdominal cavity. We took advantage of the enlargement of the umbilical orifice performed previously to exteriorize the uterus, to exteriorize the ileal loop in a less traumatic way and thus perform the procedure safely and with a lower risk of complications.

In addition to the scars, the major disadvantages of ileal segment vaginoplasty include the need of patient immobilization for a better adaptation of the flap to the tissue of the rectovaginal space, allowing the re-epithelization of the intestinal mucosa and the development of neovascularization. Therefore, patients usually receive a recommendation to stay in bed for a week.

There are rare significant complications in the immediate postoperative period of the vaginal reconstruction technique with bowel that deserve discussion. Some immediate complications are perforation of the rectum and bladder, perineal wound hematomas or necrosis of the intestinal flap, whereas late complications include stenosis, prolapse bladder, rectum or vaginal fistulae and necrosis of the urethra caused by the dilator.³²

To avoid these complications, excessive dissection of the bladder-rectal space must be avoided, and preoperative intestinal preparation, experience in intestinal surgery and meticulous suturing are essential.

Patients with surgical indication should be referred to

specialized teams, because few surgeons have extensive experience in neovagina construction and surgery by a trained gynecologist, in order to achieve a successful result.

CONCLUSION

The use of an ileal segment without a vascular pedicle is a successful option for patients with vaginal atresia, when performed by experienced surgical teams. Its advantages are the construction of a neovagina with adequate size and normal lubrication, with reduced long-term consequences. The creation of a neovagina with the aid of laparoscopy is an option that allows faster healing and reduces the psychological and aesthetic consequences of the surgery.

AUTHOR CONTRIBUTIONS

SS: Literature research; case analysis; writing of the manuscript.

IP, AH, AVL: Investigation; resources; case analysis; writing of the manuscript.

PROTECTION OF HUMANS AND ANIMALS

The authors declare that the procedures were followed according to the regulations established by the Clinical Research and Ethics Committee and to the Helsinki Declaration of the World Medical Association updated in 2013.

DATA CONFIDENTIALITY

The authors declare having followed the protocols in use at their working center regarding patients' data publication.

PATIENT CONSENT

Obtained.

COMPETING INTERESTS

The authors have declared that no competing interests exist.

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