



CASE REPORT

Double Approach Technique for Cervicovaginal Agenesis: A Case Report

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Submitted: October 20, 2025; **Accepted:** January 21, 2026; **Published:** February 02, 2026

DOI: <https://doi.org/10.25077/aoj.10.1.170-178.2026>

Abstract

Introduction: Cervicovaginal agenesis is rare female congenital anomalies. The patients usually present with primary amenorrhoea, cyclical abdominal pain, or primary infertility. A conservative surgical approach has been proposed to alleviate obstructive symptoms, construct, and maintain the patency of the genital tract, while still preserving fertility.

Case Report: A Ms. 14 year old was referred with primary amenorrhea and cyclical abdominal pain for 5 months. Physical examination and ultrasound revealed cervical dysgenesis with proximal vaginal dygenesis. Cervical canalization using double approach (laparotomy and transvaginally) was done to restore the normal anatomical integrity, maintain the functional cervical canal with a theoretical advantage of protection from ascending infection, and preservation of fertility.

Discussion: Obstructed menstrual blood, causing symptoms such as primary amenorrhea, could be a sign of female congenital anomalies. Hysterectomy has been the mainstay treatment for many decades, due to the risk of reoperation. Advances in surgical technique of conservative treatment marked the overall success rate up to 60% with few cases are reported to have pregnancy.

Conclusions: Double approach technique could be a feasible option for conservative surgery in cervicovaginal agenesis.

Keywords: Primary amenorrhea, cervical agenesis, vaginal agenesis, mullerian anomaly

INTRODUCTION

Cervicovaginal agenesis is a rare female genital anomaly, described as the impaired vaginal development, with partial or complete absence of cervical development. The prevalence is low, ranges from 1/80,000 to 1/100,000 births of all mullerian anomalies, and 50% of cases is also accompanied with vaginal agenesis.¹ Patients usually present with obstructive symptoms (primary amenorrhoea, cyclical abdominal pain), and impaired cervicouterine function (primary infertility, endometriosis, pelvic adhesion).^{1, 2} Conservative surgery offered advantages to relieve obstructive symptoms, with preservation of the reproductive function^{3, 4} We propose a novel technique of double approach for fertility preservation in adolescent unmarried patient.

CASE REPORT

A 14-year-old girl presented with primary amenorrhea and cyclic lower abdominal pain for five months. Secondary sexual characteristics were appropriate for age (Tanner stage M3P3). External genitalia appeared normal, with an intact hymen and a vaginal depth of approximately 7 cm on gentle probing. Rectovaginal examination revealed a normal-sized uterus and bilateral adnexal masses.



Figure 1. Physical exam. Gynecology exam: mp3p. Inspection: vulva and urethra unremarkable, seen structure of hymen, sondage 7 cm. RVT: Uterus size and shaped normal, cystic mass palpable in both adnexa, size approximately 6-7 cm, smooth anal mucosa, ampulla not collapsed, good TSA.

Transvaginal ultrasonography demonstrated hematometra, bilateral hematosalpinx, absence of a visible cervical canal, and proximal vaginal agenesis. The uterus was of normal size with preserved morphology. Based on clinical and imaging findings, the patient was diagnosed with primary amenorrhea due to cervical agenesis with proximal vaginal agenesis, complicated by hematometra and bilateral hematosalpinx. According to the European Society of Human Reproduction and Embryology (ESHRE) and the European Society for Gynaecological Endoscopy (ESGE) classification, the anomaly was classified as C4-V4.

Ultrasound revealed accumulation of complex fluid in intralumen of uterus, accompanied by tortuous dilatation of both tubes. No visible cervical canal and agenesis of the proximal vagina. Hematometra sized 6.55 x 3.28 x 4.42 cm, volume 49.72 cm³, right hematosalpinx sized 6.73 x 6.53 x 3.06 cm, volume 70.182 cm³ and left hematosalpinx sized 6.86 x 4.28 x 4.05 cm. volume 62.28 cm³ (Fig. 2).

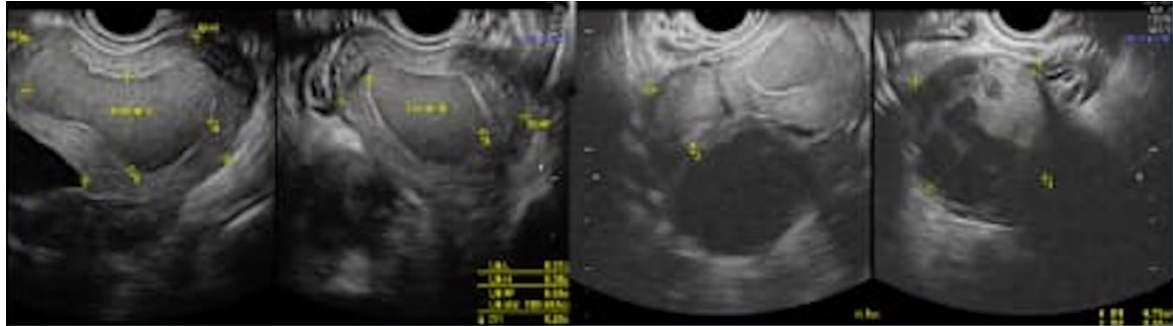


Figure 2. Radiological findings. Uterus sized 8.27 x 6.38 x 6.65 cm with volume 180.85 cm³. Cervical structure not seen. Hematometra sized 6.55 x 3.28 x 4.42 cm, volume 49.72 cm³. Right hematosalpinx sized 6.73 x 6.53 x 3.06 cm, volume 70.182 cm³. Left hematosalpinx sized 6.86 x 4.28 x 4.05 cm, volume 62.28 cm³.

Patient was then assessed with primary amenorrhea due to cervical and proximal vaginal agenesis, hematometra and bilateral hematosalpinx. Cervical canalization using double approach (laparotomy and transvaginally) was done to restore the normal anatomical integrity, maintain the functional cervical canal with a theoretical advantage of protection from ascending infection, and preservation of fertility (Fig. 3.).

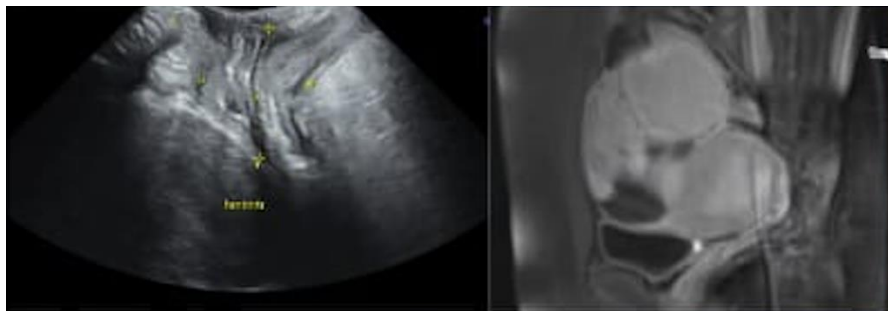


Figure 3. Radiological findings. No visible cervical canal and Agenesis of proximal vagina (left picture). Accumulation of complex fluid in intralumen of uterus is accompanied by tortuous and bilateral hematosalpinx, suggestive of cervical agenesis and proximal vaginal (right picture).

Outlines the surgical, beginning with layer-by-layer opening of abdominal wall, which revealed an enlarged uterus. During procedure, adhesiolysis was performed to release adhesions between the omentum and fallopian tubes, the intervention proceeded concluding with a uterine incision to facilitate canalization. Step-by-presented in figure 4.

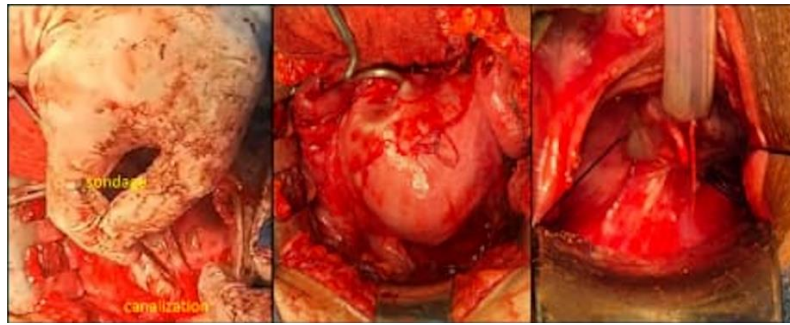


Figure 4. Treatment and technique. The abdominal wall was opened layer by layer. Following perineal incision, the operative field was exposed, revealing an enlarged uterus. Adhesiolysis was performed to release adhesions between the omentum and fallopian tubes. A right-sided endometriotic cyst measuring approximately 8 × 9 cm was identified, while the left ovary appeared within normal limits. Both fallopian tubes demonstrated findings consistent with bilateral hematosalpinx. Treatment continued with adhesiolysis followed by salpingostomy, cyst reduction, and insertion of Surgicel for hemostasis. An incision was made on the uterus to facilitate canalization.

Figure 5 illustrates the reconstructive technique used to address the cervical dysgenesis. The procedure involved a double approach where an incision was carefully made between surgical clamps to visualize successfully establishing a connection.

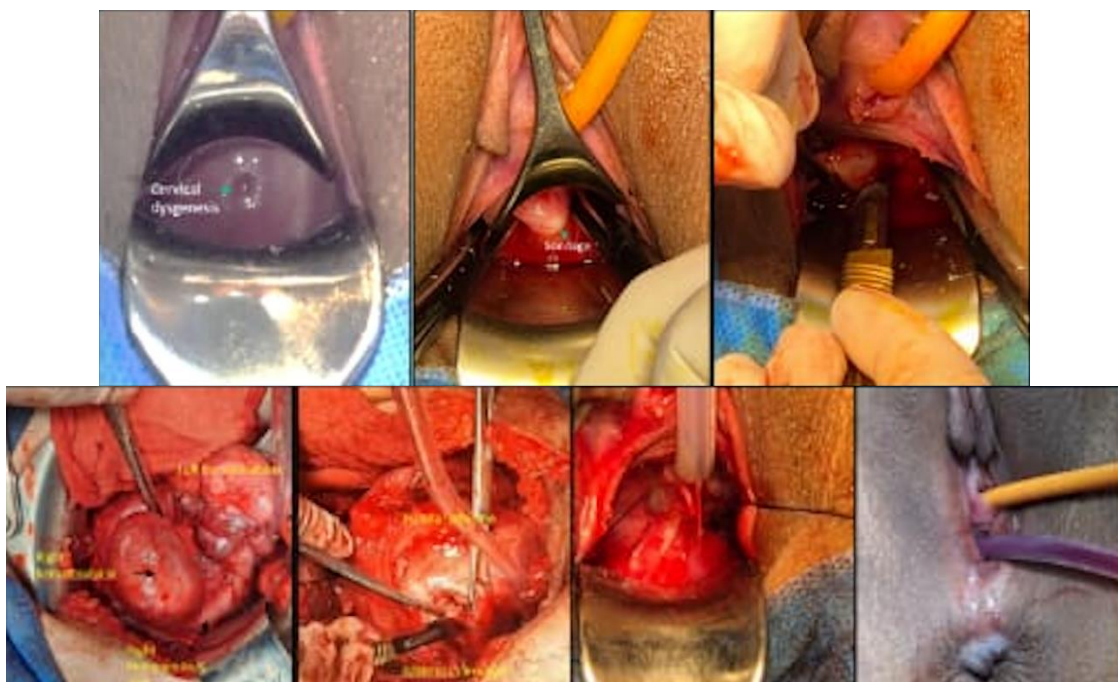


Figure 5. Treatment and double approach technique. Upon exploration, the cervix was noted to be without a cervical canal. An incision was made between clamps until the uterine sondage could be visualized from the intrauterine side. The sondage was then advanced from the uterine cavity through the newly created canal toward the vaginal introitus. A silicone catheter was inserted through this tract, and the balloon was inflated with 10 mL of sterile distilled water to maintain patency. The position of the catheter was subsequently confirmed, and the abdominal wall was closed in layers.

The table is included to benchmark and validate the findings of this specific case against broader medical literature. By comparing this patient's condition with three major historical studies. The table provides statistical evidence that the severe damage seen in this patient's body fits a well-established pattern.

Tabel 1. Associated upper tract findings in recent studies.

	Defferages (2001)	Fedele (2008)	Kriplami (2012)
Patient number (n)	18	12	14
Hematosalpinx	11 %	83 %	14.2 %
Endometriosis I, II	22 %	58 %	28.5 %
Endometriosis III, IV	22 %	-	35.7 %
Adhesions	22 %	83 %	28.5 %
Vaginal agenesis	39 %	17 %	65 %
Cervical agenesis	Not reported	83 %	64.2 %
Cervical remnant/dysgenesis	Not reported	17 %	35.7 %

DISCUSSION

Cervicovaginal agenesis a rare and rare and complex^{14,15,16} Mullerian anomaly that typically presents during adolescence with obstructive symptoms caused by a functional endometrium and an absent or non-patent genital outflow tract. Delayed diagnosis¹⁷ often results in progressive complications,²⁴ including hematometra, hematosalpinx, endometriosis, pelvic adhesions, and potential impairment of pelvic floor integrity.^{18,19} From a urogynecological perspective, prolongs retrograde menstruation and chronic pelvic inflammation may compromise surrounding structures, increasing the risk of long-term pelvic morbidity.^{20, 21}

European Society of Human Reproduction and Embryology (ESHRE) and the European Society for Gynaecological Endoscopy (ESGE) has categorized female genital anomaly. Cervical anomaly can be classified into C0 (normal cervix), C1 (septate cervix), C2 (double normal cervix), C3 (unilateral cervical aplasia), and C4 (cervical aplasia), while vaginal anomaly can be classified into V0 (normal vagina), V1 (longitudinal non-obstructing vaginal septum), V2 (longitudinal obstructing vaginal septum), V3 (transverse vaginal septum and/or imperforate hymen), and V4 (vaginal aplasia).⁵ This patient was classified as C4-V4 according to ESHRE and ESGE categorization of female genital anomaly.

Accurate diagnosis relies on combination of clinical assessment and imaging. Clinical examination is essential for confirming normal external genitalia and secondary sexual characteristics but is insufficient to evaluate internal cervical anatomy or associated pelvic pathology.²²

Clinical manifestations of cervicovaginal agenesis are primarily obstructive symptoms (primary amenorrhea, hematometra, hematocolpos, or cyclic abdominal pain) because the endometrium remains functional, while the genital outflow tract is impaired, leading to the

accumulation and obstruction of menstrual blood.⁶ This condition can further hampered cervicouterine function and may result in endometriosis, pelvic adhesion, ascending infection, and infertility.⁷ Approximately 56% of the patients with cervical agenesis are associated with pelvic endometriosis. Furthermore, patients with a delay of more than one year from the onset of symptoms to surgery had a significantly higher incidence of endometriosis ($p = 0.006$). Early diagnosis and timely surgical treatment are essential to prevent the long-term complication of this condition.⁸ Diagnosis is based on clinical evaluation and imaging findings. *Magnetic Imaging Resonance* (MRI) is the gold standard diagnostic modality; however, ultrasonography serves as the standard initial modality as it is more cost-effective and accessible. Current recommendations suggest the use 2D US combined with gynecological examination for symptomatic patients, whereas 3D ultrasound are preferred for asymptomatic cases.⁹

Due to the rarity of the condition and the limited number of studies, there is no consensus on which technique is the best. Hysterectomy was the classic initial treatment option, preferred because of the minimal postoperative complications. However, psychological issues often arise, given that this condition primarily affects adolescent women, thus, surrogacy may be required for women who have undergone hysterectomy.^{1, 10} Conservative surgery offers treatment to relieve the symptoms, restore cervicovaginal function, and preserve fertility. Conservative surgical techniques described in the literature are uterovaginal canalization or coring and drilling technique (CDT) and utero-vaginal anastomosis (UVA).^{1, 11} UVA was the earliest described conservative technique, performed by dissecting the uterovesical and rectouterine pouch, resecting the poorly developed cervix, and suturing the uterus to the high vaginal mucosa. The principle of CDT is to open the cervical canal with various instruments, drainage the content, and connecting the cervix with vagina through vaginal approach.¹¹ Canalization done in a transvaginally approach resulted in a false route and the intrusion into the uterovesical space.¹² We proposed a double approach technique (transvaginally and laparotomy) with the advantages of clearer visual surgical field. Connecting the cervix and the vagina can be done with the help through laparotomic approach by forcing a trocar down through the cervix into the vagina.¹¹

Conservative surgery demonstrates an overall success rate of approximately 60%, and only a limited number of cases have been reported to achieve subsequent pregnancy.⁷ Nevertheless, this approach is associated with a considerable risk of adverse outcomes, including endometritis, pelvic inflammatory disease, persistent pelvic pain, bowel and bladder injury, recurrent obstruction, and anastomotic stenosis, some of which may necessitate reoperation. In rare instances, procedure-related mortality has also been documented.^{12, 13}

CONCLUSION

Cervicovaginal agenesis is a rare female genital anomaly that may benefit from conservative surgical management. Double approach technique could be a feasible option for conservative surgery in cervicovaginal agenesis as it provides enhanced visualization and improved safety.

Disclosure

The authors declared no conflicts of interest. Written consent was obtained from the patient/kin of the patient.

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