CASE REPORT

Restoration to Resolution: Case Report of a Rare 20-year Longitudinal Follow-up of Cervicovaginal Reconstruction for Agenesis

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Abstract

Aim and background: Müllerian agenesis results from abnormal embryonic development of the Müllerian ducts and may lead to partial or complete absence of the cervix, uterus, or vagina. Cervicovaginal agenesis in the presence of a functioning uterus is an exceptionally uncommon condition, occurring in approximately 1 in 80,000–1,00,000 live births, and poses significant surgical and reproductive challenges. This case report aims to document the complete reproductive journey of a patient, from successful surgical restoration to resolution.

Case description: We report the case of a 37-year-old female, P2L2, with a known history of cervicovaginal agenesis, who presented at the age of 12 years with hematometra and cyclical abdominal pain. When evaluated, the diagnosis of cervicovaginal agenesis was made. Ultrasound-guided aspiration was done thrice for symptomatic relief. In 2005, she underwent unilateral nephrectomy due to the problems arising from her ectopic right kidney. She underwent cervicovaginal reconstruction (McIndoe's technique with anastomosis) in 2006. Following the restoration of menstrual patency, she achieved two spontaneous conceptions and delivered via cesarean section. After 20 years of patency, she presented with recurrent stenosis, pyometra, and hematometra, treated twice with hysteroscopy-guided drainage in 2022 and 2025. Due to the recurrence of symptoms and completed family status, she underwent a total abdominal hysterectomy and is currently asymptomatic.

Conclusion: Surgical reconstruction in cervicovaginal agenesis can achieve menstrual patency and preserve reproductive capability. Although long-term success is possible, the risk of re-stenosis persists and may necessitate rigorous follow-up. Subsequent medical or surgical interventions will be required for resolution.

Clinical significance: This case highlights that, contrary to older literature suggesting high failure rates, long-term patency and successful obstetric outcomes are achievable with appropriate surgical reconstruction, though definitive management like hysterectomy may eventually be required.

Keywords: Case report, Cervicovaginal agenesis, Hematometra, Hysterectomy, McIndoe's vaginoplasty, Müllerian anomalies.

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Introduction

Müllerian agenesis results from the embryologic underdevelopment of the Müllerian ducts, leading to a spectrum of anomalies ranging from the absence of the uterus and vagina (Mayer-Rokitansky-Küster-Hauser syndrome) to rare segmental defects. Among these, cervicovaginal agenesis—characterized by the absence or atresia of the cervix and vagina in the presence of a functional endometrial cavity—is an extremely rare congenital anomaly, with an estimated incidence of 1 in 80,000 to 1,00,000 live births. They have associated multisystemic defects such as renal anomalies (30–40%), skeletal malformations (30–40%), auditory impairment (10–25%), and rare cardiac defects.[1] This specific condition presents as a clinical challenge because the functional endometrium responds to hormonal cycles, but the outflow obstruction leads to hematometra, severe cyclic abdominal pain, and potential endometriosis.

Historically, the management of this condition has been controversial. The American Fertility Society classifies this as a Type Ib Müllerian anomaly.[2] While the definitive management via hysterectomy eliminates the risk of sepsis and pain, it irreversibly removes reproductive potential. Conversely, conservative surgical reconstruction aims to restore patency and fertility but carries a high risk of re-stenosis and ascending infection. This article presents a unique case documenting an exceptional 20-year longitudinal follow-up of a patient who, following successful reconstruction, achieved long-term patency and two spontaneous pregnancies, before late-onset recurrent stenosis, which necessitated a definitive hysterectomy.

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Case Description

The patient's clinical journey began in 2005 when she first presented at 12 years of age with symptoms of primary amenorrhea, severe

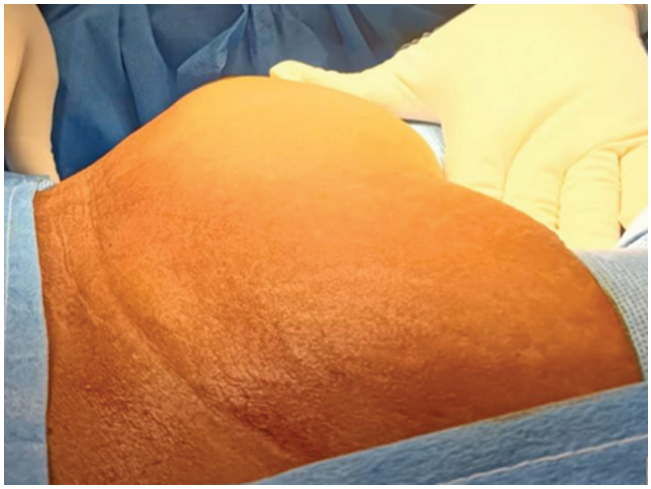
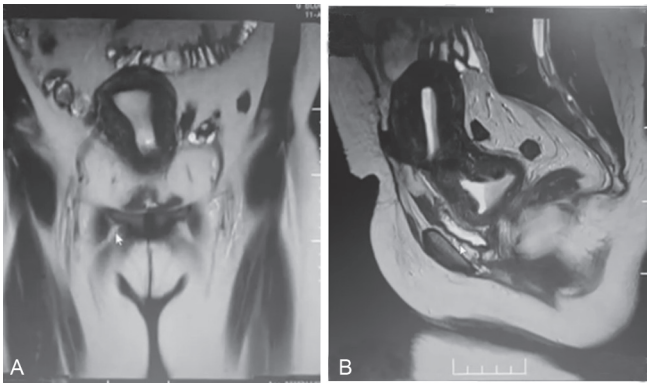
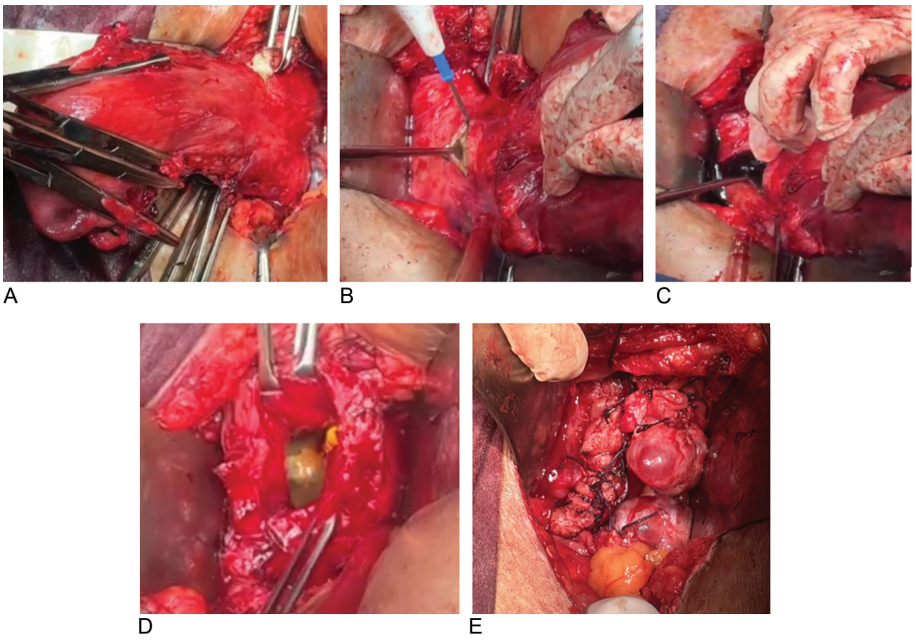


Fig. 1: Shows an abdominal mass corresponding to the size of a 24-week uterus



Figs 2A and B: (A) The uterus measuring 17.2 × 6.7 cm with a distended endometrial cavity; (B) A distinct hour-glass constriction in the mid-segment of the uterus and the endometrial cavity distended with fluid both above and below the constriction, measuring around 153cc and 294cc, respectively



Figs 3A to E: Intraoperative findings. (A) Absence of uterosacral and Mackendrot ligaments; (B) An incision made on the distended lower segment; (C) Hematocolpos drained; (D) Foley's bulb kept in the vaginal vault to drain the secretions; (E) Closure of the vaginal vault, with bilateral ovaries retained

cyclical abdominal pain, urinary retention, incontinence, and abdominal distention. Initial imaging revealed hematometra associated with cervicovaginal agenesis. Additionally, an ectopic right kidney was identified—a known association with Müllerian anomalies—which necessitated a right nephrectomy in 2005 due to recurring urinary retention and incontinence. To manage the acute distress and provide symptomatic relief from the obstructed outflow, she underwent ultrasound-guided aspiration of the hematocolpos on three separate occasions before her definitive surgical reconstruction (Figs 1 to 4).

In 2006, a decision was made to attempt uterine preservation. We successfully performed a cervicovaginal reconstruction utilizing McIndoe's technique for vaginoplasty, combined with a cervicovaginal anastomosis via an abdominoperineal approach. To maintain patency during the critical healing phase, a Silicone Malecot catheter was kept in situ for three weeks. Seven months after this initial surgery, the patient experienced a re-obstruction. This was managed by placing a Malecot catheter for an additional 3 weeks under broad-spectrum antibiotic coverage to ensure the tract remained patent. Following this, a vaginal mold was placed for 3 years to prevent vaginal stenosis. The patient subsequently resumed regular menstrual cycles and was managed on oral contraceptive pills (OCPs) to regulate flow. The patient married in 2011 at the age of 18 years. Contrary to the poor reproductive prognosis often associated with this condition, she achieved two spontaneous conceptions in 2012 and 2017. Both pregnancies

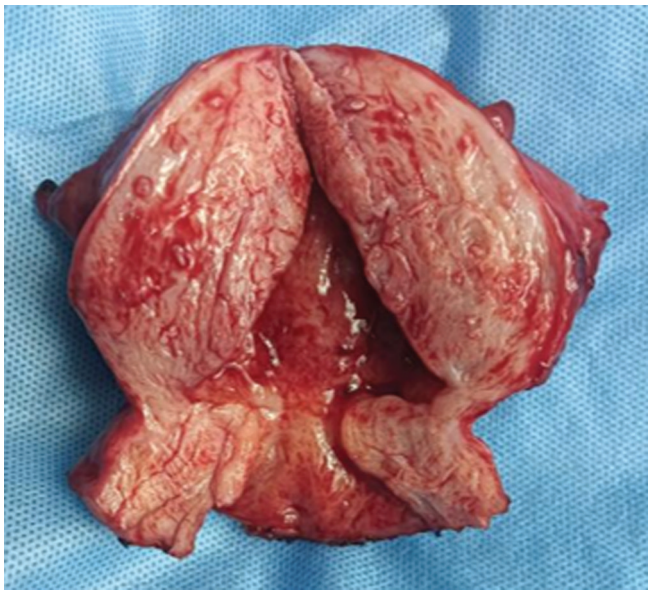


Fig. 4: The cut section of the specimen post-operatively

culminated in uneventful Cesarean sections. Intraoperatively, the os was dilated with Hegar dilators, and an intracavitary Foley catheter was placed to aid drainage and prevent immediate postpartum stenosis. This was removed after 6 days, and the patient had no complaints postoperatively.

After maintaining patency and remaining asymptomatic for 20 years, the patient developed chronic pelvic pain and signs of obstruction. She presented at age 37 with a primary complaint of severe, excruciating lower backache persisting for three weeks. She reported a change in her menstrual pattern, characterized by scanty bleeding and associated foul-smelling vaginal discharge, raising suspicion of pyometra or outflow obstruction. Imaging confirmed the diagnosis of pyometra with hematometra. Management was initially conservative; she underwent hysteroscopy-guided drainage in 2022 and 2025. However, it was complicated by re-stenosis of the reconstructed pathway. Histopathological examination at this stage revealed chronic endometritis. Transvaginal sonography and Magnetic Resonance Imaging revealed a uterus measuring 17.2 × 6.7 cm with a distinct hourglass constriction in the mid-segment. The endometrial cavity was distended with fluid both above (~153 cc) and below (~294 cc) the constriction. Given the recurrence of severe hematometra, the high risk of ascending infection, and that the patient had completed her family, the decision was made to proceed with definitive management via a total abdominal hysterectomy.

The definitive surgery presented significant technical challenges due to the distorted anatomy from previous reconstructions and congenital defects. Intraoperatively, we noted the absence of key anatomical landmarks, specifically the uterosacral and Mackenrodt's ligaments. The lower segment of the uterus was ballooned out, distorting the relationship between the ureters and the bladder. The structures had to be clamped in a modified order: round ligament, ovarian ligament, uterine vessels, and finally the tissues of the reconstructed upper vagina. A cut section of the specimen clearly demonstrated the fibrous constriction ring in the lower uterine segment responsible for the obstruction.

Discussion

The management of congenital cervicovaginal agenesis is an enduring clinical dilemma that necessitates a coordinated, multidisciplinary approach. This case illustrates the comprehensive "3R" framework: Restoration, reproduction, and resolution. While traditional management often favored definitive surgery, recent advancements include laparoscopic techniques to address these complex malformations.[1] According to the American Fertility Society, this specific defect is categorized as a Type Ib Müllerian anomaly.[2] It originates from embryologic underdevelopment of the Müllerian duct, which can manifest as atresia of the cervix, vagina, or uterus.[3] Current literature emphasizes that managing these congenital cervical malformations requires deep knowledge of anatomical variations to prevent complications.[4]

Restoration and Long-term Patency

The primary objective of surgical restoration is to create a functional outflow tract for menstruation. However, postoperative re-stenosis remains a significant hurdle. Historical reviews of congenital cervical atresia highlight that despite varied surgical attempts, long-term success is often elusive.[5] While Samantaray and Mohapatra[6] recently documented a case where patency was maintained for 18 months, our patient's outcome is exceptional, demonstrating a patent tract for 20 years following reconstruction. This longevity validates the efficacy of McIndoe's technique combined with an abdominoperineal anastomosis when supported by diligent postoperative care, including long-term molding and catheterization.

Reproductive Potential

Pregnancy in women with this condition is exceedingly rare due to the absence of cervical mucus, which is essential for sperm transport, and the mechanical barriers created by the reconstructed tract. Our patient's achievement of two spontaneous conceptions and live births reflects the rare clinical successes reported by Banu et al.,[7] proving that natural fertility is attainable with a functional uterus and a patent canal.

Resolution and Hysterectomy

Despite successful reproduction, the natural history of surgically corrected cervical agenesis often leads toward eventual obstruction. A seminal review of 30 cases managed by a common protocol showed that re-obstruction occurred in all patients over time. The resolution phase of our case—total hysterectomy after two decades—aligns with these findings, where 25 out of 30 patients ultimately required definitive surgery.[8] In this context, the hysterectomy should not be viewed as a failure of the initial restoration, but as a planned definitive step after the patient has completed her family. The late recurrence of stenosis and the development of pyometra in the patient's late 30s emphasize the critical need for lifelong surveillance.

Conclusion

Appropriate surgical intervention followed by diligent, long-term clinical surveillance enables patients to navigate the transition from anatomical restoration to successful reproduction before eventually reaching definitive resolution, thereby ensuring the opportunity for a complete reproductive life despite the challenges of complex congenital anomalies.

Clinical Significance

Cervicovaginal re-anastomosis is a viable option for patients desiring fertility, capable of sustaining patency for decades. However, patients must be counseled regarding the high likelihood of late-onset stenosis. This case supports a management algorithm that prioritizes reconstruction in adolescence to preserve fertility, while recognizing hysterectomy as the appropriate definitive management once reproductive goals are met or complications recur.

Declarations

Patient Consent for Publication

Written informed consent was obtained from the patient for publication of clinical details and images.

Artificial Intelligence (AI) Use Declaration Statement

No AI tools were used in the preparation of this manuscript.

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Nil.

Authors' Contributions (CRediT Format)

Conceptualization: Vasanthalakshmi N Gonabaktula; Investigation: Vasanthalakshmi N Gonabaktula; Supervision: Vasanthalakshmi N Gonabaktula; Writing – review and editing: Shravya Aswath; Writing – original draft: Shravya Aswath; Visualization: Shravya Aswath; Project administration: Shravya Aswath.

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