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**Case Report**

# Turning obstruction into opportunity: successful obstetric outcome in a rare and complex case of OHVIRA syndrome variant

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## ABSTRACT

Obstructed hemivagina and ipsilateral renal anomaly (OHVIRA) syndrome, also known as Herlyn–Werner–Wunderlich syndrome, is a rare Müllerian duct malformation characterized by uterine didelphys, obstructed hemivagina, and ipsilateral renal agenesis. Although rare, it bears significant gynaecological and reproductive consequences. We report a case of 24-year-old nulligravida who presented to us with chronic pelvic pain, dysmenorrhea, and persistent foul-smelling vaginal discharge. Magnetic resonance imaging demonstrated uterine didelphys with a distended obstructed left hemivagina and absent left kidney, turning clinical suspicion into reality. She underwent examination under anaesthesia followed by an exploratory laparotomy with drainage of hematopyometra, partial excision of the transverse vaginal septum, left endometrioma drainage, and left tubal delinking. Postoperative recovery was uneventful, with complete resolution of symptoms and restoration of vaginal patency. Ten months later, she conceived spontaneously, with the pregnancy localized to the right uterine horn which was meticulously monitored. At 32 weeks and 6 days of gestation, spontaneous preterm labour ensued, necessitating an emergency lower segment caesarean section. A live male neonate was delivered, and both maternal and neonatal outcomes were favourable. This case highlights the indispensable role of advanced imaging in precise anatomical delineation and emphasizes that timely, individualized surgical correction can restore reproductive potential. Despite increased risks of preterm birth and operative delivery, successful spontaneous conception and live birth are achievable in carefully managed women with OHVIRA syndrome. Early recognition remains crucial to prevent infection, endometriosis, and long-term reproductive morbidity in affected women.

**Keywords:** Müllerian anomalies, Dysmenorrhea, Subfertility, Renal agenesis, High risk obstetrics, OHVIRA syndrome

## INTRODUCTION

Müllerian duct anomalies encompass a heterogeneous group of congenital malformations resulting from impaired embryological fusion or canalization of the paramesonephric ducts. They are estimated to affect 0.1-3.8% women of reproductive age group, with variable impact on menstruation, fertility, and obstetric outcomes.[1] Among them, obstructed hemivagina and ipsilateral renal anomaly (OHVIRA) syndrome defined by uterine didelphys, obstructed hemivagina, and ipsilateral renal agenesis—represents a particularly rare and complex entity with an incidence estimated between 0.1-10% of all Müllerian anomalies.[2]

The pathogenesis of OHVIRA is attributed to the developmental failure of Müllerian and Wolffian ducts, leading to concurrent anomalies in the genitourinary systems. The clinical presentation is variable and largely depends on the degree of obstruction. Most patients become symptomatic soon after menarche, whereas some patients may present in adulthood often with infertility or recurrent pregnancy loss.

The diagnostic challenge lies in the overlap of symptoms with more common conditions like endometriosis, pelvic inflammatory disease, or obstructive vaginal anomalies. Early recognition is essential to prevent complications and long-term reproductive morbidity. Magnetic resonance imaging (MRI) remains the gold standard for diagnosis, providing excellent delineation of uterine and renal morphology.

Surgical correction that is usually achieved by excision of the vaginal septum, and drainage of hematocolpos/hematometra are the mainstay of management; restoring normal menstrual and sexual function. Successful surgical management is often associated with favorable reproductive outcomes although there remains an increased risk of preterm birth, malpresentation, and caesarean delivery.

Given its rarity and diverse clinical spectrum, each reported case of OHVIRA contributes valuable insights into optimizing timely diagnosis, individualized surgical management, and reproductive counseling.

Here we present a case of OHVIRA syndrome in a young woman who underwent corrective surgery and later achieved spontaneous conception, resulting in preterm live birth.

## CASE REPORT

A 24-year-old nulligravida presented to our OPD in February 2024 with foul-smelling vaginal discharge and intermittent dull lower abdominal pain since menarche at 13 years with regular cycles, moderate flow, and dysmenorrhea. Her initial evaluation was done at a private clinic 1 year ago where abdominal and pelvic imaging confirmed uterine didelphys, obstructed left hemivagina distended with hematocolpos, and ipsilateral renal agenesis, consistent with OHVIRA syndrome. There she underwent hysteroscopy and attempted left vaginal septal resection, and her symptoms remained unresolved.

On examination, external genitalia were normal. Per speculum evaluation revealed a blind left vaginal pouch on the left lateral aspect with copious malodorous purulent discharge. Bimanual examination suggested left-sided uterine enlargement with tenderness. Laboratory evaluation showed leukocytosis, suggestive of infection.

Ultrasonography of whole abdomen and pelvis suggested absence of the left kidney and uterine diadelphys with a collection of 7.3×4.1×5.2 cm in the lower uterine segment of the left uterine horn, cervix and upper part of the vagina. To further delineate the anatomy, CT scan and MRI of the whole abdomen and pelvis were sequentially done. The findings were in favor of Herlyn-Werner-Wunderlich Syndrome with duplicated uterine horn, cervical canal and vagina with retained fluid signal intensity within the left sided cavity suggestive of transverse septum along left paramedian component with a collection. Crucially, the left kidney was absent, confirming ipsilateral (left) renal agenesis. These findings corroborated the diagnosis of OHVIRA syndrome.

On 10 May 2024, the patient was taken up for examination under anesthesia and USG guided paracervical collection drainage. However, as the collection was organized and was difficult to access vaginally, a decision for a laparotomy was taken to manage the case.

On 21 May 2024, with the assistance of plastic surgeons, exploratory laparotomy was performed. Intraoperatively, two distinct uterine horn were seen connected along the medial aspect and the left fallopian tube and endometriotic ovary were adhered to the posterior aspect of the uterus. The right tube and ovary were normal. On palpation posteriorly and inferiorly, a bulge of 3×3 cm was felt in the left fornix. It was pushed further down and the contents aspirated (around 20 cc purulent discharge) vaginally. Left sided tubal delinking was done and the left endometrioma was drained. A small rent of 1×1 cm was created on the fundus of the left uterine horn and the partial transverse septal resection was done and the cavity was distended by the bulb of a foley's catheter for a period of 3 weeks.

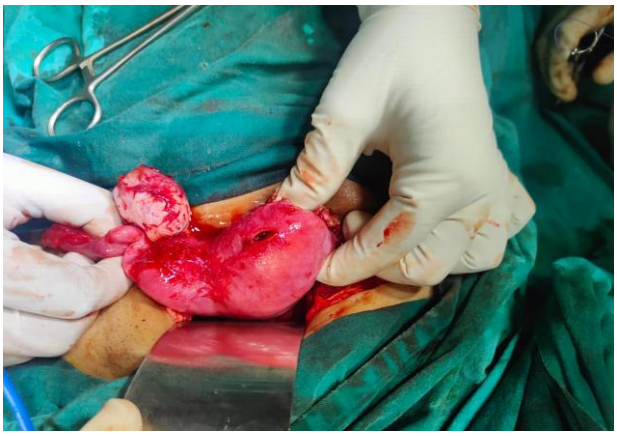

**Figure 1: Intraoperative finding showing the rent created over the left uterine horn.**

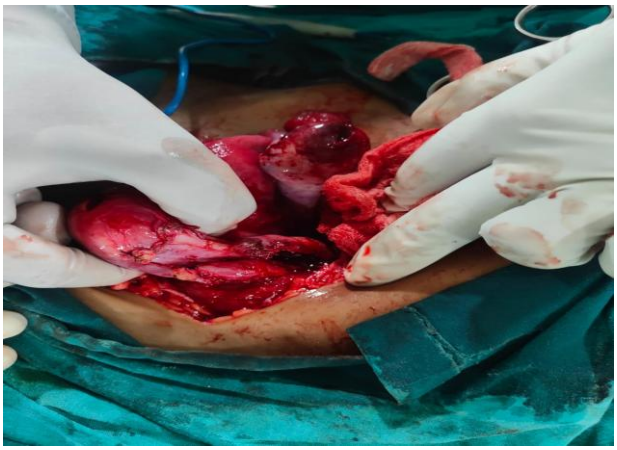

**Figure 2: Intraoperative finding showing adhesions and endometrioma.**

Post-operative care involved the administration of intravenous antibiotics along with adequate analgesia, and stringent perineal hygiene. The patient was advised abstinence for a period of 3 months to facilitate optimal healing. Combined oral contraceptive pills were prescribed for 3 months to suppress menstruation and reduce inflammation. During follow-up visits, the patient's symptoms resolved, and clinical examination revealed a patent vaginal canal without any discharge/stenosis. 10 months postoperatively, she conceived spontaneously, and USG obstetrics confirmed a single live intrauterine pregnancy localized to the right uterine horn. The antepartum period was closely monitored. Multidisciplinary management was advocated.

At 32 weeks and 6 days of gestation, the patient was admitted to the labor ward with preterm labor. Given the complex uterine anatomy, prior surgery and fetal considerations, a lower segment caesarean section was performed. A live male neonate weighing 1.7 kg was delivered. Both mother and child had an uneventful postpartum course. The neonate was admitted to the neonatal intensive care unit for supportive management.

## DISCUSSION

Purslow, in 1922, first reported this syndrome of obstructed hemivagina and ipsilateral renal anomaly.[3] Subsequently, in 1983, Herlyn and Werner recognised similar cases analogous to the anomaly and since then the anomaly has been termed as "Herlyn-Werner-Wunderlich" syndrome.[4] To aid in easy communication of the syndrome, in 2007, Smith and Laufer proposed the acronym of OHVIRA.[5] OHVIRA syndrome represents a fascinating convergence of embryological missteps involving the Müllerian and Wolffian ducts, resulting in the characteristic triad of uterine diadelphys, obstructed hemivagina, and ipsilateral renal agenesis.

OHVIRA is a type III anomaly according to the AFS and ESHRE/ESGE classifications pertaining to the new clinical and embryological classification of female genital tract malformations. It is noteworthy that as per the previous Acién classification of 1992, OHVIRA belonged to class II.[6]

The incredible rarity and variable clinical presentations of OHVIRA often lead to delayed or missed diagnoses, with many patients first presenting after years of dysmenorrhea, chronic pelvic pain, infertility, or abnormal vaginal discharge. In our patient, the presence of foul-smelling vaginal discharge, dysmenorrhea and pelvic discomfort prompted further examination and investigations culminating in the diagnosis. This underscores the importance of heightened clinical suspicion, particularly in women with infertility and prolonged, intractable dysmenorrhea.

MRI remains the gold standard for diagnosis, offering superior delineation of uterine and renal anomalies as compared to ultrasound. MRI is superior due to its multiplanar capability, high soft tissue contrast and a better characterization of the pelvic anatomy. MR imaging enables more detailed depiction of the uterus didelphys by demonstrating two separate uteri, two separate cervices along with two hemivaginas.[7] In our case, MRI played a pivotal role in correctly identifying the anomalies so that the surgery could be planned accordingly.

Surgical correction is the cornerstone for the management of such patients that is aimed at preserving fertility and relieving symptoms like dysmenorrhea, and persistent vaginal discharge. Techniques vary from simple transvaginal septal resection to more extensive procedures combining hematometra drainage, septal resection, and cervical canalization. In addition, particularly in young girls, hysteroscopic excision of the septum under transabdominal ultrasound guidance may be performed to preserve hymenal integrity.[8] Our patient benefitted from a comprehensive approach, which ensured both relief of symptoms and restoration of anatomical continuity.

Fertility outcomes following surgical correction are promising but not without challenges. Reports suggest that women with OHVIRA syndrome remain at an increased risk of miscarriage, preterm birth, malpositions and malpresentations, and caesarean deliveries.

In our patient, spontaneous conception within a year of surgery and subsequent live birth reinforces the potential for successful fertility restoration when timely interventions are undertaken.

Ultimately, this case demonstrates not only the feasibility of spontaneous conception after surgical correction but also the importance of lifelong follow up. Long term reproductive counseling, monitoring for restenosis, and careful obstetric surveillance are essential for optimizing outcomes in women with OHVIRA syndrome.

## CONCLUSION

OHVIRA syndrome, represents a rare and but clinically significant Müllerian anomaly. This case highlights the pivotal role of advanced imaging in early recognition and the effectiveness of tailored surgical correction in restoring normal anatomy. Our patient's spontaneous conception and successful live birth reaffirm that fertility potential can be preserved even in such complex anomalies when multidisciplinary management is incorporated. Beyond its rarity, this case underscores a broader message – timely intervention, individualized surgical planning, and vigilant obstetric follow up can transform a challenging anomaly into a story of a successful pregnancy outcome.

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