International Journal of Reproduction, Contraception, Obstetrics and Gynecology
Saber N et al. Int J Reprod Contracept Obstet Gynecol. 2026 Feb;15(2):725-727
www.ijrcog.org
pISSN 2320-1770 | eISSN 2320-1789

DOI: https://dx.doi.org/10.18203/2320-1770.ijrcog20260198

Case Report

Obstructed hemivagina with uterus didelphys and ipsilateral renal anomaly syndrome: a rare presentation with pyocolpos

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Received: 09 December 2025
Accepted: 12 January 2026

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ABSTRACT

Uterus didelphys with obstructed hemivagina and ipsilateral renal agenesis (OHVIRA syndrome) is a rare congenital anomaly of the female genital tract. It typically presents after menarche with progressive pelvic pain due to hematocolpos formation, leading to dysmenorrhea and a pelvic mass. We report the first documented Lebanese case of OHVIRA syndrome, involving an 18-year-old woman who presented with recurrent pelvic pain particularly during menstruation irregular cycles, and yellowish vaginal discharge. Pelvic ultrasonography, computed tomography, and magnetic resonance imaging confirmed the diagnosis. The distinct rarity of this case lies in the presence of pyocolpos, an uncommoun complication. The patient underwent surgical excision of the obstructing vaginal septum, with drainage of approximately 100 ml of purulent fluid. A Penrose drain was placed within the obstructed hemivagina the first reported use of this technique in Lebanon to ensure continuous drainage and prevent postoperative re-obstruction. The patient experienced complete symptom relief following the procedure.

Keywords: OHVIRA syndrome, Uterus didelphys, Obstructed hemivagina, Renal agenesis, Pyocolpos

INTRODUCTION

Müllerian duct anomalies constitute a spectrum of congenital disorders resulting from defective formation, fusion, or resorption of the paired Müllerian ducts during fetal development. These ducts give rise to the uterus, fallopian tubes, and the upper portion of the vagina; therefore, any developmental disturbance may lead to structural abnormalities of the female reproductive tract. Uterine anomalies occur in approximately 5.5% of the general female population.[1] One form of Müllerian duct anomalies is the uterus didelphys, which is characterized by the complete duplication of the uterine cavities, each with its own cervix and, in some cases, a separate vaginal canal. Each uterine cavity is typically associated with its own fallopian tube and ovary.[2] Obstructed hemivagina with uterus didelphys and ipsilateral renal anomaly (OHVIRA syndrome) is a rare congenital malformation of the reproductive and urinary systems. It was first described by Herlyn and Werner in 1971, who noted the link between renal agenesis and an ipsilateral blind hemivagina. Wunderlich later expanded the definition in 1976 to include uterine anomalies, establishing the term Herlyn–Werner–Wunderlich (HWW) syndrome.[3] The estimated incidence of OHVIRA syndrome is approximately 1 in 20,000 females.[4] The most common clinical manifestations of OHVIRA syndrome include pelvic pain, dysmenorrhea, a palpable lower abdominal mass due to hematocolpos and/or hematometra, and recurrent urinary

tract infections.[4] Few sporadic cases have been reported, highlighting its rarity and the limited data on optimal management.

CASE REPORT

We present a case of an 18-year-old Lebanese woman, presented to our clinic with recurrent pelvic pain, predominantly during menses, and irregular menstrual cycles. She reported menarche at age 11 and complained of yellowish vaginal discharge, but denied fever, chills, prior surgeries, or sexual activity. Speculum examination was deferred due to virginity. Ultrasound revealed two distinct hemiuteri, with a normal empty left uterus and a right vaginal cystic mass suggestive of hematocolpos. Computed tomography (CT) confirmed a single left kidney and corresponding findings (Figure 1a).

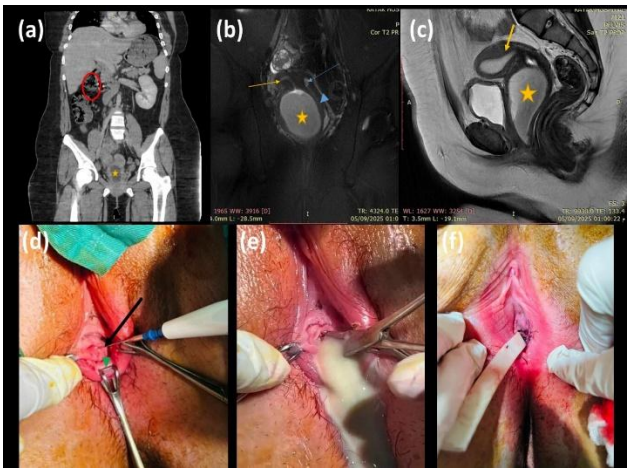

Figure 1: Radiologic and clinical findings in a case of OHVIRA syndrome presenting with pyocolpos, (a) coronal CT scan of the abdomen and pelvis showing absence of the right kidney (red circle) and a didelphic uterus with a dilated right hemivagina filled with fluid (pyocolpos) (asterisk), (b) coronal T2-weighted MR image of the pelvis demonstrates a didelphic uterus with an obstructed right uterine horn (yellow arrow) and dilatation of the right vaginal canal with fluid (asterisk) compressing the adjacent left hemivagina (blue arrowhead). Note the normal left uterine horn (blue arrow), (c) sagittal T2-weighted MRI scan showing a didelphys uterus with a right hemiuterus (arrow) which communicates with an obstructed and fluid-filled right hemivagina (asterisk), (d) obstructed right hemivagina (black arrow) with intact hymen (green arrowhead), (e) drainage of the pyocolpos, and (f) intraoperative image: vaginoplasty and marsupialization of the vaginal edges.

MRI further delineated the pelvic anatomy, demonstrating duplication of the uterine cavity and cervix with a vertical vaginal septum. The right uterine horn measured 9 cm and showed cystic dilation of the right vagina and cervix, with high T2 signal and low-level internal echoes, consistent with hematometrocolpos or pyocolpos (Figures 1b and c). These findings were diagnostic of OHVIRA syndrome. To the best of our knowledge, this is the first reported case of OHVIRA syndrome in a Lebanese patient. Under general anesthesia, the patient's hymeneal orifice appeared normal (Figure 1d). The right vaginal septum was excised, resulting in drainage of approximately 100 ml of purulent fluid (Figure 1e). Marsupialization of the vaginal edges was performed, and a Penrose drain was placed into the right uterine cavity through the new opening to ensure complete drainage and prevent postoperative re-obstruction or fluid recurrence (Figure 1f).

Culture grew out sensitive *Escherichia coli*.

Postoperatively, she experienced complete resolution of her symptoms.

DISCUSSION

Obstructive Müllerian anomalies should be considered in adolescents with recurrent pelvic pain or dysmenorrhea, even with normal menstruation, as obstruction may be unilateral. Early diagnosis is crucial to prevent potential long-term reproductive complications (i.e. pelvic infection, endometriosis, and infertility).[5]

Ultrasound and magnetic resonance imaging (MRI) are the preferred imaging modalities for diagnosing and planning the management of OHVIRA. Although ultrasound can detect the anomaly, MRI provides superior assessment of uterine morphology and relationships with adjacent structures, owing to its multiplanar capabilities and broader field of view.

Despite its high sensitivity, MRI was unable to detect the purulent nature of the collection in our case. This is partly explained by low clinical suspicion, as the patient did not report typical signs of infection such as fever or chills. Therefore, an infected hematocolpos should be considered even in the absence of systemic manifestations of infection.[6]

A thorough clinical assessment combined with ultrasound and MRI is usually sufficient for preoperative diagnosis and planning of OHVIRA syndrome. In the absence of tubal or pelvic pathology, surgeons can proceed directly to vaginal septum excision, with the approach tailored to the size and definition of the septal bulge. The standard technique involves complete septal excision with mucosal edge approximation or marsupialization, as in our case, to ensure adequate drainage and prevent postoperative re-obstruction. Hysteroscopy may be used selectively, while laparoscopy is reserved for complications such as hematosalpinx requiring salpingectomy.[6-8]

CONCLUSION

OHVIRA syndrome should be suspected in adolescents with cyclic pelvic pain, a vaginal mass, and unilateral renal agenesis. MRI is the key diagnostic tool, accurately

defining uterine anatomy, renal anomalies, and the nature of any obstructed vaginal collection. Early diagnosis enables timely surgical intervention, preventing long-term complications. Laparoscopy is generally reserved for associated tubal or pelvic pathology rather than primary diagnosis.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Saber N, Barakat F, Awala M, Nakhel R, Saber M, Barakat H. Obstructed hemivagina with uterus didelphys and ipsilateral renal anomaly syndrome: a rare presentation with pyocolpos. Int J Reprod Contracept Obstet Gynecol 2026;15:725-7.