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Case Report

Obstructed hemivagina and ipsilateral renal anomaly syndrome in an adolescent female – diagnostic challenges and surgical management: a case report

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ABSTRACT

Obstructed hemivagina and ipsilateral renal anomaly (OHVIRA) syndrome, also known as Herlyn–Werner–Wunderlich syndrome, is a congenital Müllerian anomaly characterized by uterine duplication, obstructed hemivagina and an ipsilateral renal anomaly. It commonly presents after menarche with cyclic pelvic pain and dysmenorrhea due to retained menstrual blood. A 14-year-old girl presented four months after menarche with lower abdominal pain and dysmenorrhea. Ultrasonography demonstrated uterine duplication, hematometrocolpos and an abnormal left kidney, suggesting OHVIRA syndrome. Magnetic resonance imaging (MRI), however, suggested a unicornuate uterus with a non-communicating rudimentary horn and did not clearly demonstrate the vaginal obstruction, creating a diagnostic dilemma. Examination under anaesthesia and diagnostic laparoscopy demonstrated two non-communicating uterine horns and an obstructed hemivagina. Excision of the obstructing vaginal septum and drainage of approximately 100 mL of retained blood were performed. At six-month follow-up, the patient had regular menstrual cycles and complete resolution of pelvic pain. Discordant imaging findings may complicate the diagnosis of OHVIRA syndrome. Correlation of clinical findings with ultrasonography and MRI, followed by examination under anaesthesia and laparoscopy, when necessary, can establish the diagnosis. Timely vaginal septum excision relieves obstruction and may help prevent long-term complications.

Keywords: OHVIRA syndrome, Herlyn–Werner–Wunderlich syndrome, Uterine didelphys, Obstructed hemivagina, Renal agenesis

INTRODUCTION

Obstructed hemivagina and ipsilateral renal anomaly (OHVIRA) syndrome, also known as Herlyn–Werner–Wunderlich syndrome, is a rare congenital Müllerian anomaly characterized by uterine duplication, unilateral vaginal obstruction and an ipsilateral renal anomaly.[1,2] It results from abnormal development of the Müllerian and mesonephric ducts and may remain clinically silent until menarche.[3,4] Patients commonly present with progressive dysmenorrhea, cyclic pelvic pain or a pelvic mass caused by hematocolpos and/or hematometra.[1,5] Because menstrual flow may continue through the unobstructed hemivagina, diagnosis can be delayed, increasing the risk of endometriosis, pelvic adhesions, infection and impaired fertility.[6,8]

Ultrasonography is commonly the initial imaging modality, whereas MRI provides detailed delineation of Müllerian and vaginal anatomy.[4,5] The usual treatment is surgical excision of the obstructing vaginal septum to restore drainage and preserve reproductive potential.[7,9]

CASE REPORT

A 14-year-old female presented with low-grade lower abdominal pain for two months, associated with nausea and vomiting. She had attained menarche four months earlier and reported dysmenorrhea with irregular menstrual cycles lasting 3–5 days at intervals of 45–50 days. Her vital signs were stable and hemoglobin was 12 g/dL. Abdominal examination revealed mild lower abdominal tenderness without guarding, rigidity or a palpable mass.

Imaging findings

Pelvic ultrasonography revealed uterine duplication. The right uterine horn measured 4 × 2 cm and had a normal endometrial cavity without fluid collection. The left uterine horn measured 6 × 3 cm and showed a 5 × 2 cm hypoechoic intra-cavitary collection with internal moving echoes and no vascularity, with myometrial thinning of approximately 7 mm. A hematometrocolpos measuring approximately 7 × 6 cm was also noted. The left kidney appeared atrophic, suggesting an ipsilateral renal anomaly. These findings were suggestive of OHVIRA syndrome. Pelvic MRI demonstrated an enlarged uterus with significant hematometra and ipsilateral hematosalpinx. A non-communicating rudimentary horn containing functional endometrium was reported, with no right-sided hematometra or hematosalpinx. The vagina was not clearly visualized. The left kidney was not visualized, with compensatory hypertrophy of the contralateral kidney. The MRI findings therefore raised the possibility of a unicornuate uterus with a non-communicating rudimentary horn rather than classic OHVIRA syndrome. Because of the discordance between ultrasonography and MRI, examination under anaesthesia and diagnostic laparoscopy were planned, with definitive surgical treatment according to the intraoperative findings.



Figure 1: Preoperative pelvic ultrasonography demonstrating uterine duplication with hematometra and hematocolpos in the obstructed hemivagina. The retained menstrual blood appears as a hypoechoic collection with internal echoes.

Surgical management

Under general anaesthesia, the patient was placed in the lithotomy position. On per-vaginal examination, the right-sided cervical os was visualized and a bulge was noted on the left vaginal wall due to accumulation of menstrual blood. Diagnostic laparoscopy revealed two non-communicating uterine horns. The right horn was normal in size, whereas the left horn was uniformly distended and measured approximately 7 × 4 cm. Both fallopian tubes appeared normal. The findings confirmed an obstructed hemivagina in the setting of uterine duplication. The obstructing vaginal septum was excised and approximately 100 ml of retained blood was drained. The septal margins were everted and sutured with 2-0 delayed-absorbable polyglactin (Vicryl) sutures. A 14-Fr Foley catheter was inserted, the balloon was inflated with 20 ml of saline and the catheter was left in situ for 48 hours to maintain patency and facilitate drainage under antibiotic coverage.



Figure 2: Follow-up pelvic ultrasonography after surgical excision of the vaginal septum showing resolution of hematometra and hematocolpos with no residual fluid collection.

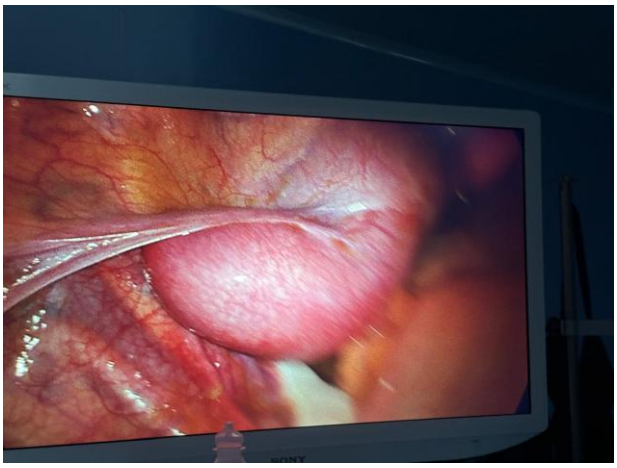


Figure 3: Diagnostic laparoscopic view demonstrating uterine duplication with a distended left uterine horn secondary to retained menstrual blood.

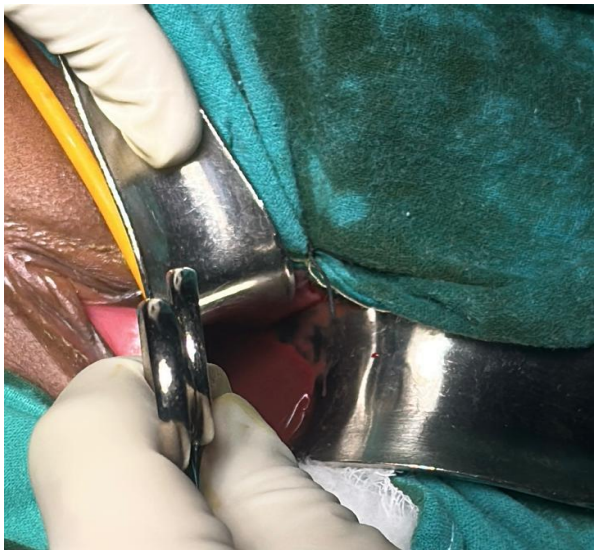


Figure 4: Intraoperative image showing incision of the obstructing vaginal septum with evacuation of retained menstrual blood from the obstructed hemivagina.

Postoperative course

Follow-up ultrasonography on postoperative day 3, after removal of the Foley catheter, showed no residual collection. The patient was advised regular follow-up with annual surveillance. At six-month follow-up, she reported regular menstrual cycles every 30–32 days, lasting 5–6 days, with complete resolution of pelvic pain.

DISCUSSION

OHVIRA syndrome is a congenital anomaly involving the Müllerian and mesonephric duct systems. The clinical phenotype is typically characterized by uterine duplication, unilateral obstructed hemivagina and an ipsilateral renal anomaly.[1,3,4] Clinical manifestations usually become apparent after menarche because menstrual blood accumulates in the obstructed hemivagina and ipsilateral uterine cavity, producing hematocolpos, hematometra and, in some patients, hematosalpinx.[1,5] Progressive dysmenorrhea and cyclic pelvic pain are common presenting symptoms, but diagnosis may be delayed when menstruation continues through the unobstructed hemivagina.[6]

The present case is notable for the diagnostic discrepancy between ultrasonography and MRI. Ultrasonography demonstrated uterine duplication, hematometrocolpos and a left renal abnormality, findings that suggested OHVIRA syndrome. MRI, in contrast, reported a possible unicornuate uterus with a non-communicating rudimentary horn and did not clearly visualize the vagina. Distortion of the pelvic anatomy by retained blood and difficulty delineating the vaginal septum can complicate interpretation of Müllerian anomalies. MRI remains an important modality for anatomical delineation, but imaging findings should be interpreted in conjunction with the clinical examination and other imaging findings.

In the present patient, definitive characterization was achieved through examination under anaesthesia and diagnostic laparoscopy. The identification of two non-communicating uterine horns, together with the obstructed hemivagina and left renal anomaly, supported the final diagnosis of OHVIRA syndrome. Published literature similarly emphasizes the value of correlating clinical, radiological and operative findings when the anatomy is atypical or the imaging findings are discordant.[5,9,10] Renal anomalies are a frequent component of Müllerian duct anomalies, and recognition of a unilateral renal anomaly should prompt consideration of associated genital tract abnormalities.[3,4] Prolonged obstruction can lead to endometriosis, pelvic adhesions, infection and adverse reproductive outcomes, making early diagnosis and treatment important.[6,8,11]

Excision of the obstructing vaginal septum is the standard conservative surgical treatment for OHVIRA syndrome. It relieves the obstruction, allows drainage of retained menstrual blood and preserves the uterus and reproductive potential.[7,9] In this patient, septal excision and drainage resulted in resolution of the obstructive symptoms, with restoration of regular menstrual cycles at six-month follow-up.

The main clinical lesson from this case is that a discordant MRI report should not exclude OHVIRA syndrome when the clinical presentation and ultrasonography are suggestive. Careful vaginal examination and, in selected cases, diagnostic laparoscopy can resolve uncertainty and allow definitive treatment during the same procedure.

CONCLUSION

OHVIRA syndrome should be considered in adolescent girls presenting after menarche with cyclic pelvic pain or dysmenorrhea, particularly when a renal anomaly is present. Although MRI is valuable for delineating Müllerian anatomy, discordant or inconclusive imaging may occur. Correlation of clinical examination with imaging findings and, when necessary, examination under anaesthesia and laparoscopy can establish the diagnosis. Timely excision of the obstructing vaginal septum can relieve symptoms and may reduce the risk of long-term complications.

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