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Dr. Deepika Theresa T
M.S., OBGYN, F.MAS, Minimal
Access Surgery, Division of
Gynecologic Endoscopy, Sri
Ramachandra Medical Centre,
Chennai, Tamil Nadu, India

Dr. Usha Rani G
M.D, OBGYN, Professor, Division
of Gynecologic Endoscopy Sri
Ramachandra Medical Centre
Chennai, Tamil Nadu, India

Virtual reality aided minimal access surgery in the management of OHVIRA syndrome: A case series

Deepika Theresa T and Usha Rani G

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Abstract

OHVIRA syndrome also known as Herlyn Werner Wunderlich syndrome is an extremely rare mullerian anomaly. The incidence is very small. Only isolated cases have been recorded and published. OHVIRA represents Obstructive HemiVagina and Ipsilateral Renal Agenesis (OHVIRA). It presents with a triad of obstructive vaginal septum, ipsilateral renal anomalies and various types of uterine malformations. Symptoms usually begin post menarche. Patients present with cyclical abdominal pain or severe dysmenorrhea, chronic pelvic pain or prolonged spotting. This is due to the underlying obstruction by the vaginal septum causing hematocolpos and hematometra. Although 3D ultrasound and MRI are excellent modalities for diagnosis of OHVIRA syndrome, we intend to emphasise on the added advantages of upcoming technology such as 3D virtual reality reconstruction, which helps to plan our surgery precisely. We present here, 2 cases of OHVIRA syndrome with different presentations, diagnosis aided by radiological imaging and immersive virtual reality and managed by minimal invasive surgery. We also aim to discuss here, different theories in its pathophysiology and different techniques in the hysteroscopic management of OHVIRA syndrome.

Keywords: OHVIRA syndrome, chronic pelvic pain, virtual reality, hysteroscopy, Herlyn Werner Wunderlich syndrome

Introduction

Normal embryology of the uterus and cervix involves fusion of the paired paramesonephric ducts, and lower vagina from the sinovaginal bulb. Any deviation from this, resulted in various mullerian anomalies of the uterus, cervix and the vagina, as classified by ESHRE.

The syndrome involves a triad of ipsilateral renal agenesis, obstructive vaginal septum and associated uterine anomalies. Patients present with chronic pelvic pain, abnormal uterine bleeding or subfertility.

3D Ultrasound and MRI are the mainstay for diagnosis. 3D immersive virtual reality adds on to the depths of the presenting problem, and also helps in precisely planning the surgery. We present two rare cases of the syndrome, where 3D virtual reality aided our diagnosis and management.

Case reports

Author has patient and guardian consent for publication

Our first case is a 14 year old teenage girl, who attained menarche at 10 years of age, presented with severe dysmenorrhea, chronic pelvic pain and prolonged bleeding during menses. On general examination, she had normal Body mass Index, gait, spine, breasts, thyroid and external genitalia. She was further evaluated with blood parameters such as Complete blood count, Thyroid function test, Prolactin, Follicle stimulating hormone, Luteinising hormone, Anti mullerian hormone and were found to be within normal limits. Ultrasound abdomen and pelvis was done, found to have right renal agenesis and possible septate uterus and hematocolpos was observed. Proceeded with an MRI pelvis which showed a complete septate uterus (2cm in the upper part and 1-2mm in the lower uterus and cervix). Hematocolpos was observed of size 4 x 4 x 3.2cm involving the whole of the vaginal cavity (Figure 1A). No hematometra noted. Likely suggestive of imperforate hymen or OHVIRA syndrome.

Hence evaluation was further proceeded by incorporating the MRI images into our immersive 3D virtual reality system software, in which a complete septate uterus was confirmed. Right obstructive hemivagina with oblique septum in the vagina noted. No hematometra (Figure 1B). Additional points from the Virtual reality were the exact septum length and thickness (Figure 1C) and we were able to virtually perform the surgery beforehand in the software, which was very beneficial and time saving during the actual surgery. Right renal agenesis present. Left kidney and ureter normal. Diagnosis of OHVIRA syndrome was confirmed. According to the ESHRE classification, it was U2b C0 V2.

Corresponding Author:
Dr. Deepika Theresa T
M.S., OBGYN, F.MAS, Minimal
Access Surgery, Division of
Gynecologic Endoscopy, Sri
Ramachandra Medical Centre,
Chennai, Tamil Nadu, India

We decided to proceed with hysterolaparoscopy followed by hysteroscopic resection of the vaginal and uterine septum. Intra operatively, in laparoscopy, the external contour of the uterus was normal. Broad fundus noted. Bilateral tubes and ovaries were normal (Figure 1D). Rest of the peritoneal cavity was normal. Vaginoscopy was done. Oblique vaginal septum till the introitus with bulging right hematocolpos noted. Small fistulous opening in the vaginal septum at the cervicovaginal junction noted. Proceeded with resection of the vaginal septum using resectoscope of 22F with collin knife (monopolar) and glycine as distension medium (Figure 1E). Chocolate coloured fluid drained out as we resected the vaginal septum. Resection was completed. Further, resectoscope introduced into the uterus and 2mm thin septum in the cervix noted. Same excised. Complete uterine septum noted, same resected with collin knife until bilateral ostia was visualised from the level of internal os. Silicone catheter was introduced into the cavity and distended with 50cc distilled water and removed after 48 hours on post-operative day 2. The patient was comfortable and was discharged. The patient was started on Oral contraceptive pills, continued for 3 cycles, she was followed up after 3 months with Ultrasound. She made a complete recovery with normal menstrual cycle.

Our second case is a 12 year old girl who attained menarche at 11 years of age presented with prolonged spotting during cycles. Patient had a history of laparoscopic right nephrectomy secondary to right renal dysplasia. Clinical examination was done and normal. Blood parameters were normal. Ultrasound abdomen and pelvis done which showed uterine didelphys with mild hematocolpos. MRI pelvis was done and findings were consistent with OHVIRA syndrome-separate divergent uterine horns, uniformly separate uterine cavities with no evidence of communication in between (Figure 2A). Complete division noted till the external OS. Two cervices noted. Two separate upper vagina noted (secondary to the high oblique vaginal septum) and single lower vagina noted. Small 2.6 x 2cm right

hematocolpos noted. Small ovarian cyst of 2 x 2 cm in the left side. Again, evaluation was proceeded with immersive 3D virtual reality in which didelphic uteri and right upper hematocolpos was visualised (Figure 2B). According to ESHRE, it was U3b C2 v2. Additional findings studied in virtual reality in this patient were the presence of adhesions due to previous surgery, abdominal wall thickness and thereby port positions was planned in detail.

We proceeded with hysterolaparoscopy. Entry done through palmaris point. Mild adhesions released through sharp dissection through harmonic scalpel. Intra operatively in laparoscopy, didelphic uterus noted. Adnexa normal. Vaginoscopy done, left cervix visualised. No obvious bulge from the hematocolpos noted. Small 5mm opening noted in the medial side of the left cervix (Figure 2C), minimal brownish discharge seen through the opening. Opening slightly enlarged with hysteroscopic biopsy forceps after which the right cervix was also visualised. Proceeded with vaginal septum resection (around 1-2cm). Patient was comfortable and was discharged after 48 hours. She was followed up after 3 months with Ultrasound, and made complete recovery with resumption of normal menstrual cycle. Unlike our first case, our second case did not require a foley bulb for adhesion prevention, as the second case had a didelphic uterus. But, in the first case, complete uterine septal resection was also accomplished for her complete separate uterus.

Discussion

In 1980, OHVIRA syndrome was introduced. Pathophysiology involves failure of one side of the mesonephros to develop, leading to malformation of the ipsilateral renal kidney or complete agenesis, which in turn leads to improper paramesonephros and hence proximal vagina abnormalities. Acien proposed a different theory contributing to OHVIRA syndrome, which proposes that there is a probability of contribution of Wolffian duct in the formation of the proximal vagina, thereby causing oblique vaginal septum in the syndrome.

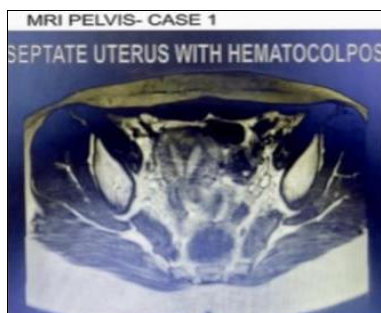


Fig 1A: MRI showing septate uterus



Fig 1B: Virtual reality creation showing single uterine exterior with hematocolpos



Fig 1C: Uterine cavity (interior) showing septum in green colour



Fig 1D: Laparoscopic findings: normal fundal contour with normal adnexa



Fig 1E: Hysteroscopy findings: Vaginal septum excised with resectoscope and chocolate-coloured fluid drained

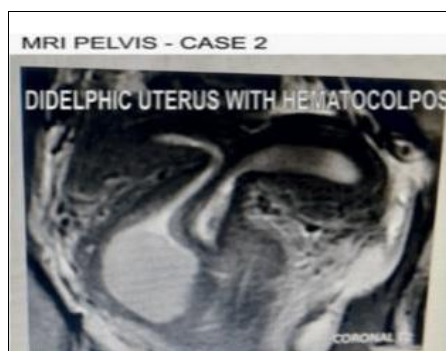


Fig 2A: MRI showing didelphic uterus with hematocolpos in right side

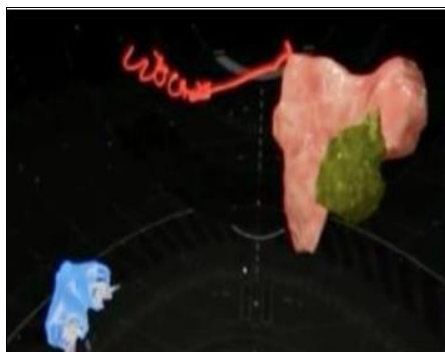


Fig 2B: Virtual reality recreation- possible fenestration in the cervix visualised



Fig 2C: Vaginoscopy findings-2-3mm fenestration adjacent to left cervix noted

Patients usually present after menarche with progressive dysmenorrhea, lower abdominal pain, a paravaginal mass, foul mucopurulent discharge and intermenstrual bleeding due to the hemi hematocolpos. There is also a risk of endometriosis, subfertility and pelvic adhesions. Diagnosis is by multi modal approach, which includes thorough history taking, clinical examination, 3D ultrasound, MRI. Hysterolaparoscopy is the gold standard.

In our case, 3D immersive virtual reality aided in diagnosis and in precisely planning our surgery. 3D immersive technology helps us visualise the organs with exact shape, size and precision from skin to skeleton. It has various modes including zoom in, zoom out, view only the organ of interest, adjacent vital structures, surgical knife technique which helps us to virtually perform the surgery pre operatively. Virtual reality helps in visualising minute details such as adhesions, blood vessels, location of ureter, etc. Hence every part of surgery such as port position, incision site, proximity of ureter and bowel in cases of endometriosis, can be precisely visualised and planned before surgery.

Management of OHVIRA syndrome involves minimal access surgery by hystero laparoscopy

In cases of the didelphic uterus, cavities are not to be disturbed. In case of a septate uterus, septal resection can be done in the same sitting or in the second sitting. As most of the patients with OHVIRA are adolescents, preserving hymen is important.

In most cases, oblique vaginal septum causing obstructive hemi vagina is identified as soon as the vaginoscope is introduced, as it bulges out. Then vaginal septum can be resected with the Collins knife or point coagulator in the most dependent part of the bulge. Usually dark chocolate colored fluid escapes out, which further guides us to proceed with vaginal septal resection. But if bulge is not identified in 3 percent patients, there may be underlying fenestration in the cervix or in the vaginal septum. In those cases, a hysteroscopic catheter is left inside the fenestration, inflated with air or mannitol and then the bulge becomes prominent and septum can be easily excised.

Post operatively, for adhesion prevention, patients require intra uterine (if uterine septum is respected) distended for 48 hours. Post Op estradiol valerate is also accepted for 1 month followed by Medroxy progesterone, which helps in re-epithelisation and adhesion prevention.

Ethics Statement

Author has patient and guardian consent for publication

This case report is sent for publication in accordance with the ethical principles by the hospital. The authors certify that they

have obtained all appropriate patient consent. The patient understands that his/her name and initial will not be published and due efforts will be made to conceal his/her identity.

Author contributions

All authors were involved in the drafting, editing and approval of the manuscript for publication. In addition to this, each author contributed the following: Dr. Deepika Theresa-collected data, reviewed articles, wrote and edited manuscript. Dr. Usha Rani-reviewed articles, edited manuscript.

Data availability statement

The data underlying this article will be shared on reasonable request to the corresponding author.

Conclusion

Hysterolaparoscopy is the main stay for management and diagnosis for OHVIRA Syndrome. 3D Immersive virtual reality is definitely a value addition for the diagnosis and management of rare and tricky cases such as these, or in endometriosis, ventrofixed uterus, large myoma, mullerian anomalies, etc.

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Not applicable

Conflicts of interest

All authors declare no conflicts of interest.

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