VIDEO ARTICLE

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A combined endoscopic and ultrasonographic approach to a complex U4a uterine anomaly

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ABSTRACT

Background: Uterine malformations are congenital anomalies arising from abnormal Müllerian duct development during embryogenesis. These can be linked to vaginal cysts, resulting in complex malformations. One rare form is the unicornuate uterus, where only one duct develops, leading to complications like severe pain due to a rudimentary, non-communicating horn.

Objectives: To describe a combined approach using ultrasound, hysteroscopy, and robotic-assisted laparoscopy for complex uterine anomalies.

Participant: A 30-year-old nulliparous woman with unilateral kidney agenesis and acute pelvic pain referred to our centre.

Intervention: 2D ultrasound suggested a complex malformation. 3D ultrasound and magnetic resonance imaging confirmed a U4a uterus. Hysteroscopy revealed a hemicavity with one tubal ostium. Robotic-assisted laparoscopy enabled right salpingectomy and removal of the rudimentary horn while preserving the ovary. Intraoperative ultrasonography guided the drainage of vaginal cysts. As a result, vaginal cysts were drained, and the rudimentary horn was removed with ovarian preservation. The patient was discharged without complications and spontaneously conceived a healthy pregnancy 8 months later.

Conclusions: Unicornuate uterus with non-communicating horn and renal agenesis is a rare condition. A combined approach using ultrasound, hysteroscopy, and robotic-assisted laparoscopy allows comprehensive evaluation and treatment.

What is New? This is the first reported case of simultaneous and synergistic use of hysteroscopy and robotic-assisted laparoscopy for complex genital malformations under ultrasonographic guidance.

Keywords: Unicornuate uterus, hysteroscopy, robotic-assisted laparoscopy, complex genital malformations, non-Müllerian anomalies

Introduction

The prevalence of unicornuate uterus is approximately 0.1% in the general female population, 0.5% in infertile women, and 2% in those with a history of miscarriage.[1] A unicornuate uterus may be associated with a rudimentary horn, which can be either communicating or non-communicating. One in 35 cases is associated with hematometra due to obstruction of a non-communicating rudimentary horn. The rudimentary horn may or may not contain functional endometrium.[2,3]

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Diagnosis of obstructive Müllerian anomalies typically occurs in adolescence due to blood retention and pain; however, mild menstrual pain may delay diagnosis.

In complex genital malformations, Müllerian anomalies can coexist with non-Müllerian anomalies due to defects in mesonephric duct remnants.[4-7]

We present the case of a 30-year-old nulliparous woman with unilateral kidney agenesis who experienced acute pelvic pain. She was diagnosed with a unicornuate uterus [European Society for Gynaecological Endoscopy/ European Society of Human Reproduction and Embryology (ESGE/ESHRE) U4a classification], featuring a non-communicating horn and associated vaginal cysts.

This case demonstrates the benefit of combining transvaginal ultrasound (2D/3D), hysteroscopy, and robotic-assisted laparoscopy for comprehensive diagnosis and treatment. Ultrasonography provided essential preoperative guidance, while hysteroscopy and laparoscopy facilitated surgical management, including the identification and drainage of vaginal cysts and removal of the rudimentary uterine horn while preserving fertility. This case is notable for being the first to treat a complex uterine anomaly using simultaneous hysteroscopy and laparoscopy under ultrasonographic guidance.

Methods

A 30-year-old nulliparous patient was referred to our centre due to the onset of severe abdominal pain. Her medical history was notable for right unilateral renal agenesis. Additionally, she reported a pattern of light menstruation interspersed with prolonged periods of amenorrhea. Both her personal and family medical histories were unremarkable. From a professional standpoint, she was a classical ballet dancer. Following a thorough clinical and laboratory evaluation, the patient underwent ultrasound assessment, initially with 2D imaging, followed by 3D ultrasound for further anatomical delineation. As an adjunct diagnostic modality, magnetic resonance imaging (MRI) of the abdomen and pelvis was also performed. The diagnostic workup was further complemented by performing an inpatient hysteroscopy using a 5-mm continuous-flow hysteroscope.

Results

Initial 2D ultrasound suggested a complex uterine malformation with a non-communicating rudimentary horn, hematometra, and vaginal cysts. 3D ultrasound and MRI confirmed a Class U4a uterus, as per the ESGE/ESHRE classification. Hysteroscopy revealed a hemicavity with a single tubal ostium. Robotic-assisted laparoscopy successfully facilitated the removal of the rudimentary horn and right salpingectomy. Retroperitoneal access allowed for direct visualization of the ureter to rule out other urological anomalies.

Intraoperative ultrasonography enabled precise identification of the vaginal cysts. The caudal vaginal cyst was drained via a minor incision using a 5Fr electrode, resulting in the release of thick, dark mucus. A separate cranial vaginal cyst was also emptied without complications. The patient was discharged the next day with no adverse events.

Eight months after the surgery, the patient spontaneously conceived and is currently carrying a healthy pregnancy.

Discussion

A unicornuate uterus with a non-communicating rudimentary horn is a rare Müllerian anomaly associated with endometriosis, pelvic pain, and infertility. In this case, Herlyn-Werner syndrome, a complex urogenital anomaly, was excluded through vaginoscopy, which confirmed the absence of an atretic hemivagina anterolateral to the patent vagina. Similarly, imaging ruled out Wunderlich syndrome, as no blind-ending hemivagina was detected. Heller[8] described how mesonephric duct developmental anomalies can lead to Gartner's duct retention, resulting in vaginal cysts, as observed in this patient.[9-11] Acién suggested that mesonephric anomalies may contribute to renal agenesis due to failed ureteral bud sprouting.[12,13] Notably, laterality was evident in our patient's anomalies, including right-sided vaginal cysts, a cavitated non-communicating rudimentary horn, and renal agenesis. The failure of the Wolffian duct's inductive function on the Müllerian duct contributes to uterine duplication and ipsilateral renal agenesis. This developmental mechanism may explain the observed laterality of the anomalies.[14] A combination of ultrasound, vaginoscopy, and robotic-assisted laparoscopy provides a comprehensive approach to diagnosing and treating complex malformations. Bermejo et al.[15] highlighted that 3D ultrasound is comparable to MRI imaging. However, we opted for MRI to rule out any additional urological anomalies.

We chose to drain the cysts due to the patient's new onset of dyspareunia. Imaging and hysteroscopy

showed that the cysts were not large enough to require excision, as noted by Thapa and Regmi[16] Based on the cysts' appearance and according to the findings from Bats et al.[17], we ruled out malignancy risk. To minimize invasiveness, particularly in a young patient, we opted for cyst drainage. This involved creating a wide opening in the cyst wall and selectively coagulating the cyst bed using a 5Fr bipolar electrode for both incision and coagulation. The endoscopic approach enhanced safety through direct visualization, while intraoperative ultrasound offered real-time guidance for identifying and draining vaginal cysts, especially for the second cyst with a more cranial development.

Pre-surgical imaging revealed poorly defined anatomical planes, raising concerns about potential access to the abdominal cavity during hysteroscopic drainage of the vaginal cysts, which were also in continuity with each other. Therefore, we opted for a robotic approach, which, in addition to the inherent advantages of laparoscopy—minimized blood loss, accelerated recovery, and next-day discharge—provides enhanced precision and control, particularly in cases with complex or unclear anatomical structures. Furthermore, the dual endoscopic approach provided definitive treatment in a single procedure, avoiding further surgeries. Moreover, the patient later achieved a spontaneous pregnancy, demonstrating the success of this multidisciplinary approach in preserving fertility.[18]

The patient sought care after experiencing her first episode of severe pelvic pain. Her prolonged amenorrhea, likely a result of the intense physical and emotional demands of her career as a professional classical ballet dancer, may explain why the condition went undetected until adulthood.

This is the first reported case of treating a complex female genital malformation using simultaneous vaginoscopy and laparoscopy under ultrasonographic guidance, presenting a promising approach for similar cases in the future.

Conclusion

A unicornuate uterus with a non-communicating rudimentary horn and ipsilateral renal agenesis represents a rare and complex clinical condition. In this case, the combination of Müllerian and non-Müllerian anomalies required a multidisciplinary approach involving 2D/3D ultrasound, hysteroscopy, and robotic-assisted laparoscopy.

This approach allowed for comprehensive evaluation and treatment, ensuring preservation of the patient's fertility and leading to a favourable outcome, including a spontaneous pregnancy. Intraoperative ultrasonography provided crucial real-time guidance, particularly for the identification and management of vaginal cysts.

To our knowledge, this is the first reported instance of simultaneous hysteroscopy and laparoscopy under ultrasonographic guidance for treating such a rare and complex malformation. This combined approach offers an effective and minimally invasive solution for managing congenital uterine anomalies and associated conditions.

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Transparency: The authors affirm that this manuscript is an honest, accurate, and transparent account of the case reported. All relevant details have been included, and no important information has been omitted. The patient's identity has been protected in accordance with ethical standards.

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Video 1. https://youtu.be/0VOispgg3cE