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Case Report

Unveiling the Hidden Horn: A Rare Case of Rudimentary Uterine Horn with Endometriotic Diseases

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#Eually Contribution

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A B S T R A C T

Background: A rudimentary uterine horn is a rare congenital anomaly that results from the abnormal development of one Müllerian duct, while the other duct remains underdeveloped. This condition is often associated with a unicornuate uterus and may lead to severe complications, such as endometriosis, due to retrograde menstruation. The diagnosis of this anomaly is challenging due to non-specific symptoms and high misdiagnosis rates.

Case summary: We present the case of a 45-year-old woman who experienced progressive dysmenorrhea for 10 years and prolonged menstrual periods for 3 months. Imaging studies, including ultrasound and MRI, suggested a double uterus with multiple fibroids and potential hydrosalpinx. During laparoscopic surgery, the patient was found to have a rudimentary uterine horn with a small connection to the unicornuate uterus, right ovarian cysts, severe pelvic adhesions, and endometriosis. The surgical procedures included the removal of the rudimentary horn, right ovarian cystectomy, and excision of fibroids from both the left and right uteri. Postoperative pathology confirmed leiomyoma and adenomyosis. The patient is currently under follow-up with normal ultrasound and laboratory findings.

Conclusion: This case highlights the importance of early recognition and accurate diagnosis of rudimentary uterine horn, which is often misdiagnosed due to atypical presentations. A combination of imaging techniques, clinical evaluation, and surgical exploration is critical for proper management. Early intervention can help alleviate symptoms, prevent complications like endometriosis, and improve patients' quality of life.

Keywords: Rudimentary uterine horn; Unicornuate uterus; Endometriosis; Congenital uterine anomaly

1. Introduction

The rudimentary uterine horn is a rare form of congenital uterine anomaly, resulting from the development of one Müllerian duct and the developmental deficiency of the contralateral Müllerian duct. It is characterized by the presence of only the uterine corpus and fallopian tube, with an absence of the cervix and vaginal structure on the affected side[1]. The unicornuate uterus accounts for approximately 5% of Müllerian duct anomalies,

and is often associated with a rudimentary uterine horn. Clinically, it can be divided into three types[1]. Manifestations of a rudimentary uterine horn is diverse and subtle[2], with some individuals remaining asymptomatic and the condition going unnoticed indefinitely. Additionally, the diagnosis of this condition is challenging and prone to misdiagnosis, which can lead to a range of complications. Non-rudimentary uterine horn pregnancy (RHP) is often misdiagnosed as endometriosis, while rudimentary uterine horn pregnancy is most likely to be mistaken for tubal pregnancies. A pregnancy in a rudimentary uterine horn can lead to complications such as uterine rupture[3], posing a risk to the patient's life. We report a case of a rudimentary uterine horn combined with a unicorous uterus and associated with endometriosis (EMT) to enhance awareness and understanding of this condition and to reduce the incidence of misdiagnosis and missed diagnosis.

2. Case presentation

2.1. Clinical and pathological data's

Patient, female, 45 years old, presented to our hospital on June 23, 2024, due to "progressive dysmenorrhea for 10 years and prolonged menstrual period for 3 months." The outpatient ultrasound suggested the possibility of a rudimentary uterine horn, a uterine fibroid on the left side, and adenomyosis on the right side **(Figure 1)**, leading to admission to the hospital. The patient has regular menstrual cycles, with a 30-day cycle, moderate flow, and severe dysmenorrhea. Last menstruation: 2024-07-05. Previous menstruation: 2024-05-24. Past medical history: diabetes and hypertension for 9 years. Obstetric history: 1-0-0-1, cesarean section in 2003.Preoperative expert ultrasound (2024-07-15): Didelphic uterus (single cervix, single cervical canal) with multiple fibroids in the left uterus, the largest measuring approximately 454437mm, one of which is submucosal, adenomyosis with fibroids in the right uterus, a small amount of fluid in the right uterine cavity, and a poorly echoic area on the right measuring 74×57×32mm, suggesting the possibility of hydrosalpinx **(Figure 2)**. On July 16, 2024, the patient's CA-125 level was measured at 279.40 U/ml, and CA 199 was 102.4 U/m. A pelvic MRI revealed no significant abnormalities in the liver, gallbladder, spleen, pancreas, and left kidney **(Figure 3)**. A urinary system ultrasound indicated a right kidney absence status **(Figure 4)**.

On July 17, 2024, the patient underwent a combined laparoscopic and hysteroscopic surgery. Intraoperative findings included: the left uterus was mid-positioned, enlarged to the size of a 2-month pregnancy, with an irregular shape. There were two myoma-like tissue protrusions on the anterior wall of the uterus, approximately 4 cm and 3 cm in diameter, mostly protruding outside the serosa, and hard in texture. There was one myoma-like tissue protrusion on the posterior wall of the uterus, about 5 cm in diameter, partially protruding outside the serosa, and hard in texture. The left ovary and fallopian tube appeared normal; the right uterus was mid-positioned, enlarged to the size of a 2-month pregnancy, with a full shape, and closely adhered to the left uterus. The right fallopian tube was thickened and tortuous, with the ampulla visible. The right ovary had a cystic mass about 6 cm in diameter. The right ovary, fallopian tube, and parts of the intestines, mesentery, and pelvic wall were closely adhered and encapsulated. Scattered brown endometrial implantation lesions were seen on the intestinal wall, about the size of rice grains, and no significant fluid was observed in the pelvic cavity. The adhesions throughout the pelvic cavity were released, the mesentery and isthmus of the right fallopian tube were transected, and the right fallopian tube was removed; the viscous chocolate-like fluid in the cystic cavity of the right ovarian tumor was aspirated, the ovarian rupture was opened along the long axis of the ovary, and the cyst wall was bluntly peeled off and completely removed. The raw surface was rinsed with physiological saline and hemostasis was achieved with bipolar electrocoagulation. The endometrial implantation lesions on the intestinal wall were excised; the adhesions between the left and right uteri were separated, the right uterine artery was clamped, and the right uterus was removed. It was observed that the two uteri were connected by a small hole with a diameter of about 2mm, and the raw surface was sutured; the anterior wall of the uterus was cut horizontally with a unipolar electro hook to the pseudo capsule of the myoma, and three uterine fibroids on the left side were peeled off, the largest being about 4×4×3cm, and the raw surface was continuously sutured with a 1/0 absorbable suture for the muscle layer and the myometrium. The right uterine body was rotated and cut with a uterine rotator, and several myoma-like tissues were observed on the cross-section of the uterus, and three uterine fibroids were similarly rotated and cut. The right ovarian cyst, right fallopian tube, right uterus, three uterine fibroids on the left side, and the intestinal wall endometriosis lesions were all sent for pathological examination. Hysteroscopic surgery was performed to remove the protruding tissue in the uterine cavity, with a total size of about 4.0×4.0×2.0cm, and a curettage was performed, and a small amount of endometrial tissue was scraped out. The uterine cavity excision and the scraped material were sent for routine pathological examination. Intraoperative findings are illustrated in **(Figure 5)**. Postoperative hysteroscopic re-examination showed no obvious space-occupying lesions or active bleeding. Intraoperative rapid pathology showed: (right ovarian tumor) benign cyst; (left uterine fibroids) leiomyoma. (Entire right uterus) ① (uterus) Leiomyoma. ② (uterus) Adenomyosis. The surgery went smoothly, with 2000ml of fluid replenished during the surgery, 500ml of blood loss, and 400ml of urine output.

Postoperative pathology indicates: endometriotic lesions on the intestinal wall. Right ovarian tumor: endometrial cyst of the ovary. Right uterus: ① Leiomyoma ② Adenomyosis. Right fallopian tube: hydrosalpinx with chronic inflammation. Left uterine fibroid: leiomyoma. Endometrial scrapings and electro-resection specimens from the left uterus: submucous leiomyoma.

Postoperative follow-up: On August 19, 2024, the patient came to our hospital's outpatient department for follow-up: CRP (panel), blood cell [five-category] analysis showed: basophil percentage 1.1%, eosinophil count 0.66×10^9/L, basophil count 0.10×10^9/L, red cell distribution width CV 15.6, platelet count 601×10^9/L, plateletcrit 0.42%. Ultrasound examination: The uterus is anteverted, measuring 50×40×32mm, with regular shape and clear contour. The myometrium of the uterus is uneven. The endometrial thickness is 6mm, uneven **(Figure 6)**. Outpatient treatment with dienogest was prescribed. Currently, regular follow-up is ongoing, and follow-up ultrasound, CRP, and blood cell tests are all normal.

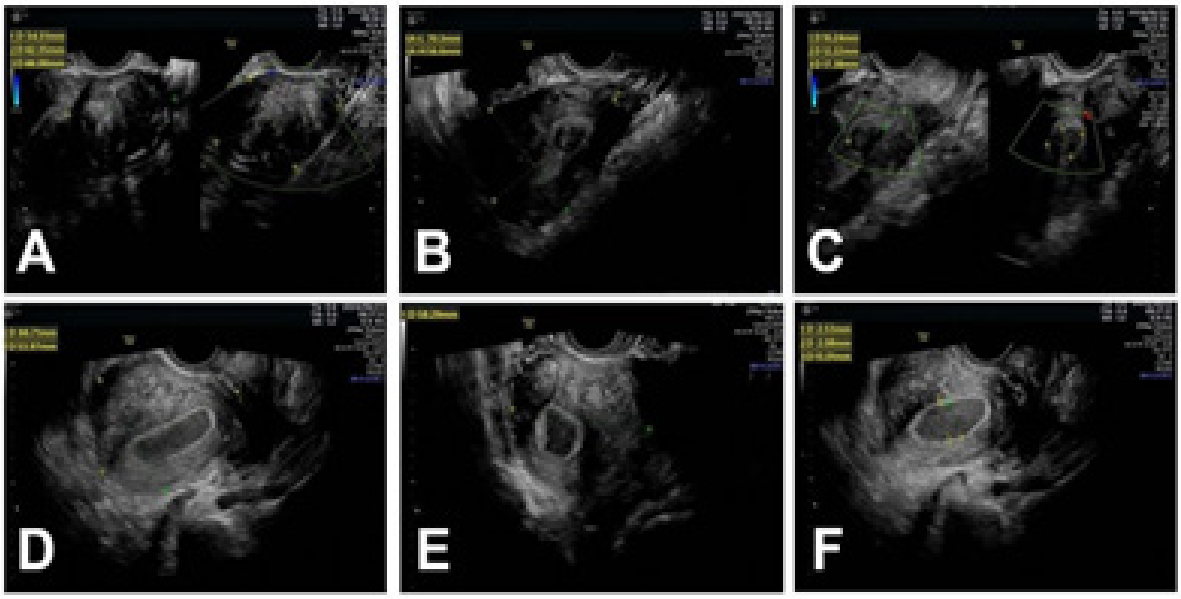

Figure 1: Outpatient ultrasound. Suggesting the possibility of a unicornuate uterus. Panels A to C are the left uterus, and panels D to F are the right uterus.

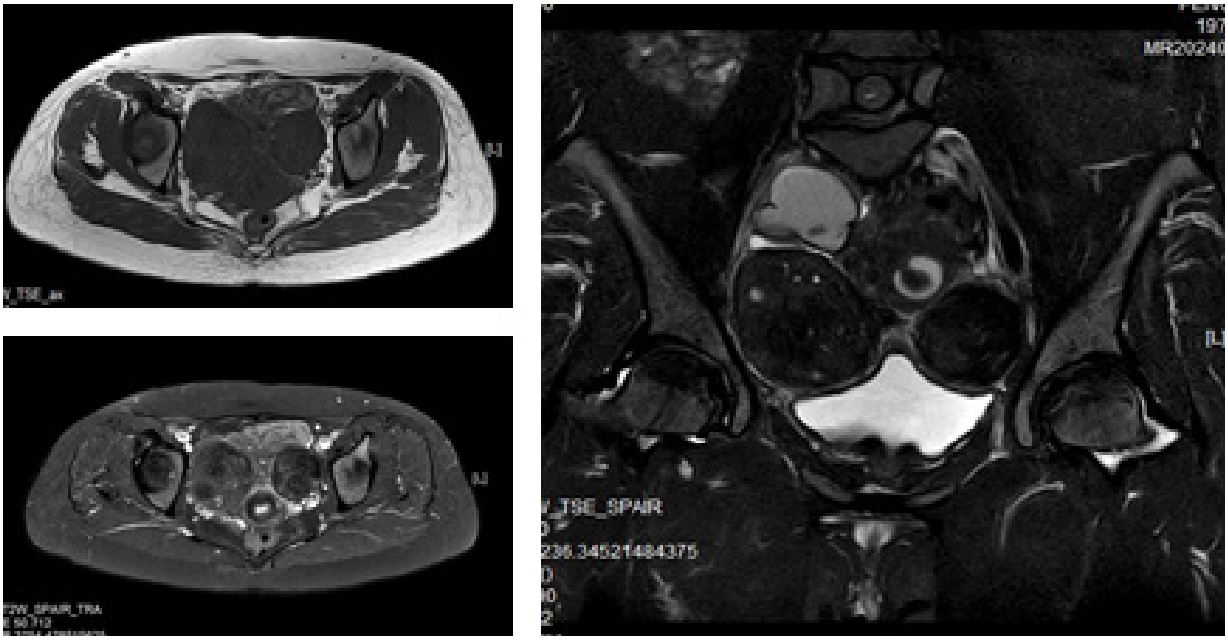

Figure 2: Expert ultrasound. The left image is the left uterus, and the right image is the right uterus.

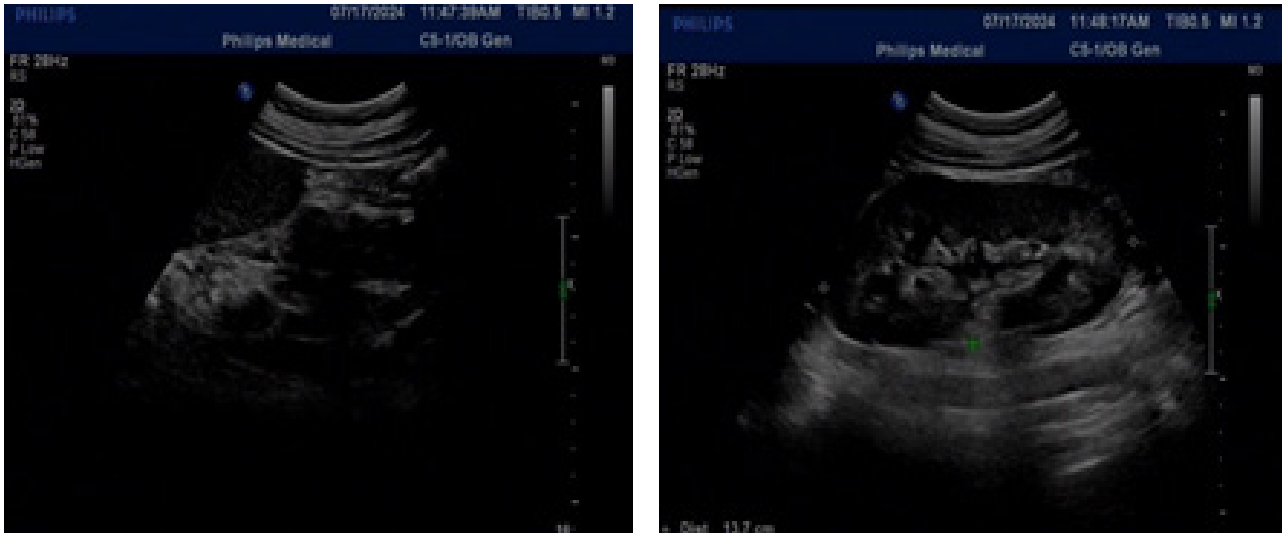

Figure 3: Pelvic MRI. No significant abnormalities observed in the liver, gallbladder, spleen, pancreas, and left kidney.

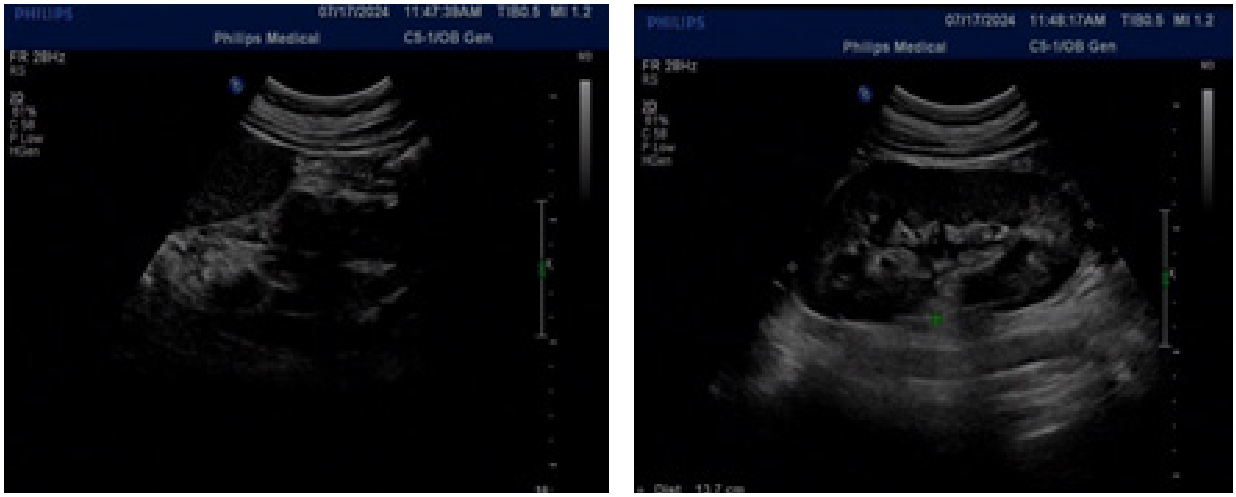

Figure 4: Urinary system ultrasound. The left image shows absence of the right kidney, and the right image shows a solitary left kidney.

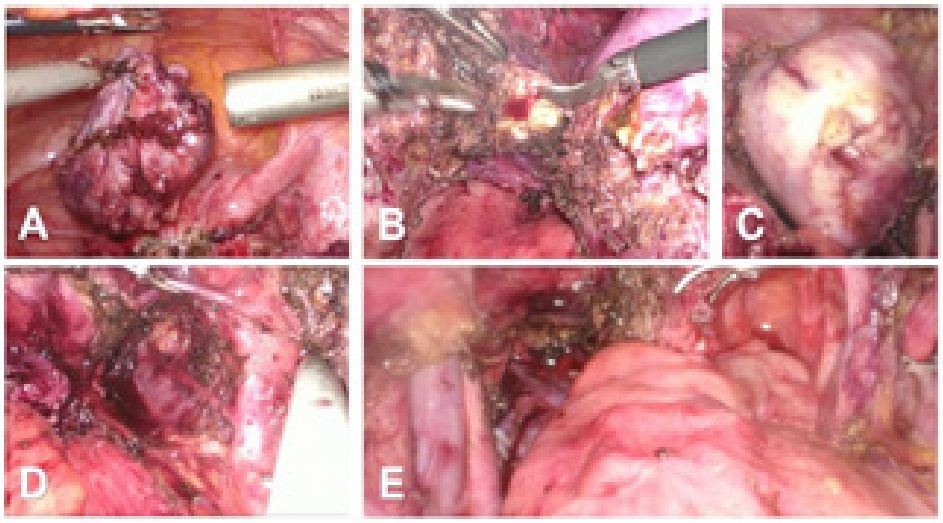

Figure 5: Intraoperative Findings.

A: Right ovarian chocolate cyst;

B: A 2mm small opening connecting the rudimentary uterus to the unicornuate uterus;

C: Uterine fibroid on one side of the unicornuate uterus;

D: Variations in branching and course of blood vessels on the rudimentary uterus side; E: Comparison of pelvic floor anatomy between the unicornuate uterus and the rudimentary uterus reveals vascular variations on the rudimentary uterus side.

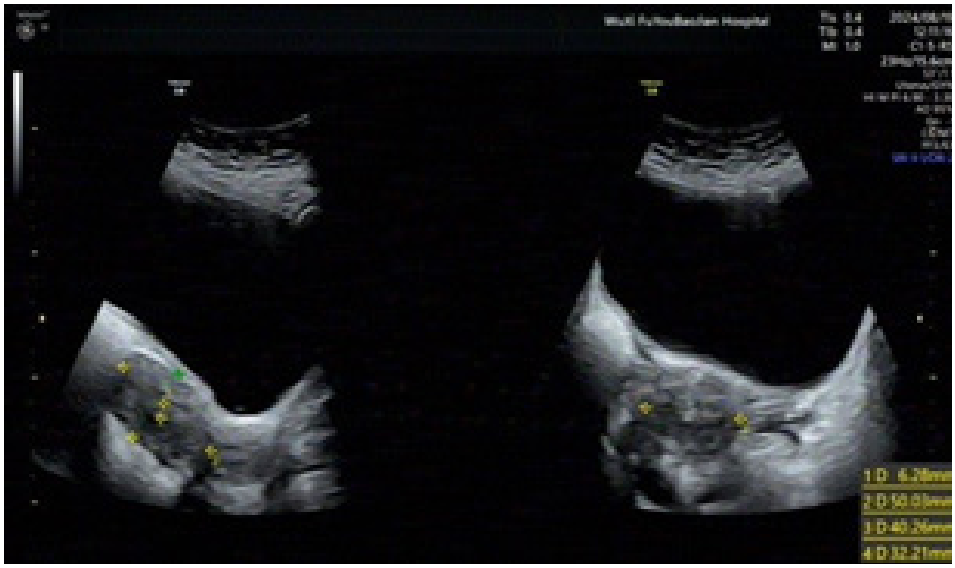

Figure 6: Follow-up ultrasound.

2.2. Diagnosis, differential diagnosis, and analysis of the causes of misdiagnosis of a rudimentary uterine horn

The incidence of rudimentary uterine horn is low, and its clinical manifestations are atypical, making misdiagnosis and missed diagnosis likely. When patients present with the following clinical manifestations, the possibility of a rudimentary uterine horn should be considered: (1) Dysmenorrhea: A major clinical manifestation of type II rudimentary uterine horn. When secondary dysmenorrhea occurs, one should be alert to uterine abnormalities. Severe dysmenorrhea may be due to retrograde menstruation in the unicornuate uterus, which does not communicate with the normal uterine cavity, leading to endometriosis and blood stasis[4]. History of recurrent miscarriages and infertility: A rudimentary uterine horn affects the patient's fertility, as the uterine muscle wall is poorly developed and cannot withstand the growth and development of the fetus, making it prone to severe pregnancy complications such as uterine rupture[5]. (3) Abdominal pain: Commonly seen in the mid to late stages of pregnancy with a rudimentary uterine horn, it may occur due to uterine rupture or torsion[6]. (4) Pelvic mass: Gynecological examination may reveal a mass closely related to the uterus, which can be mistakenly diagnosed as an ovarian tumor. (5) Signs of urinary system abnormalities: Further confirmation of the presence of urinary system deformities can be obtained through imaging examinations. In addition to clinical manifestations, auxiliary tests can also be combined to assist in the diagnosis of rudimentary uterine horn. These mainly include two-dimensional ultrasound, three-dimensional ultrasound, MRI, and hysterosalpingography (HSG). Two-dimensional ultrasound is cost-effective but cannot accurately observe the coronal section of the uterus, increase the rate of misdiagnosis and missed diagnosis of the disease. In contrast, three-dimensional ultrasound has higher sensitivity and is of great value in disease diagnosis. Studies have shown that combining three-dimensional ultrasound with routine two-dimensional ultrasound helps to improve the accuracy of disease diagnosis[7].Additionally, MRI is helpful for classifying rudimentary uterine horn, and HSG can be used to assist in the diagnosis of uterine anomalies.

The misdiagnosis rate of rudimentary uterine horn is quite high, and it needs to be differentiated from the following conditions: (1) Ovarian tumors: Both can manifest as masses in the adnexal region, connected to the uterus by a pedicle. (2) Endometriosis: A rudimentary uterine horn may lead to endometriosis due to retrograde menstruation, but endometriosis itself is not a rudimentary uterine horn, and it needs to be distinguished through clinical manifestations and imaging examinations. (3) Tubal pregnancy: Both rudimentary uterine horn pregnancy and tubal pregnancy can present with a history of amenorrhea followed by rupture and abdominal pain, and the interstitial part of the tubal pregnancy may also protrude, making it difficult to differentiate in the early stages of pregnancy. In summary, the main reasons for the misdiagnosis and missed diagnosis of rudimentary uterine horn are the lack of understanding of the condition by physicians, insufficient history taking and examination, leading to incomplete clinical data, and over-reliance on imaging examinations.

3. Discussion

According to the ASRM classification, unicornuate uterus is categorized as Type II. Clinically, this malformation group is further divided into four subtypes: Type a refers to a rudimentary uterine horn that communicates with the unicornuate uterine cavity; Type b, where the rudimentary horn has a cavity but does not communicate with the unicornuate uterus; Type c, a rudimentary horn without a cavity that is connected to the unicornuate uterus only by a fibrous band; and Typed, the absence of a rudimentary horn[8]. This anomaly results from abnormal development of one side of the Müllerian duct during embryogenesis and is frequently associated with urinary tract malformations. Statistics indicate that unicornuate uterus is accompanied by renal hypoplasia or ectopic kidney in 15% of cases, with 40% of this exhibiting congenital renal agenesis on the side of the rudimentary horn[8,9]. If the rudimentary uterine horn contains functioning endometrium, it undergoes cyclic shedding during menstruation, leading to retrograde menstrual flow and accumulation within the uterine cavity. This condition increases the likelihood of gynecological issues, such as dysmenorrhea and chronic pelvic pain, and may gradually develop into endometriosis and infertility[10]. As a rare congenital uterine anomaly, the rudimentary uterine horn is often difficult to diagnose and treat early in clinical practice due to nonspecific early symptoms and limited accuracy of imaging studies[11].

We present a rare case of a Type IIa rudimentary uterine horn malformation associated with ipsilateral renal agenesis and severe endometriotic disease. Preoperative transvaginal ultrasound suggested the possibility of a rudimentary uterine horn, while MRI indicated a double uterus. Although research shows a good correlation between MRI and surgical findings in diagnosing congenital uterine malformations[12], in this case, the small communication between the rudimentary horn and the unicornuate uterus may have led to a misdiagnosis on MRI. The patient's right kidney agenesis was a key diagnostic clue, highlighting the need for clinicians to assess patients based on clinical symptoms, comprehensive imaging studies, and cutting-edge medical knowledge to devise optimal treatment plans.

The primary goals in treating a rudimentary uterine horn are to alleviate symptoms, prevent complications, and preserve reproductive health. In this case, the patient had experienced progressively worsening dysmenorrhea over 10 years, severely affecting her quality of life, making laparoscopic resection of the rudimentary horn an appropriate choice[13].

Endometriotic disease encompasses both endometriosis and adenomyosis, conditions in which active endometrial tissue is ectopically located. Adenomyosis refers to the invasion of endometrial tissue into the myometrium, forming endometriotic lesions, while endometriosis refers to the presence of endometrial tissue outside the uterine cavity, primarily in pelvic organs and the peritoneum. The pathogenesis of endometriosis remains controversial and includes theories such as retrograde menstruation, autoimmune response, metaplasia of coelomic epithelium, and lymphovascular spread. The retrograde menstruation theory suggests that fragments of endometrial tissue containing active glands and stroma reflux through the fallopian tubes into the peritoneal cavity, where they adhere and invade the underlying mesothelium[14]. This theory aligns with epidemiological evidence linking EMs to factors such as increased menstrual bleeding, shorter cycle length, higher frequency of menstruation, and an increased incidence of reproductive tract obstructions[15]. Common complications of congenital Müllerian duct malformations include hematosalpinx, endometriosis,

chronic pelvic pain, and adhesions, all secondary to retrograde menstruation[16]. In cases where the rudimentary uterine horn does not communicate with the unicornuate uterus, the inability to expel menstrual blood often leads to endometriotic disease. In this case, the pathogenesis appears to align with the retrograde menstruation theory, as intraoperatively, a 2 mm communication between the rudimentary horn and the unicornuate uterus was identified. The outflow tract had been obstructed for years, causing progressively worsening dysmenorrhea. Fragments of menstrual blood containing active endometrial cells likely refluxed through the fallopian tubes into the ovaries and pelvic cavity, resulting in adenomyosis in the rudimentary horn and endometriotic lesions in the bowel wall and right ovary. Treatment of endometriotic disease requires a tailored approach based on the patient's age, symptoms, and reproductive desires, along with long-term postoperative management.

Once the diagnosis is established, surgical management becomes critical. In this case, we first addressed the endometriotic lesions in the bowel wall and right ovary to prevent the spread of chocolate cyst fluid in the pelvic cavity. Next, we proceeded with the resection of the rudimentary uterine horn. Given that rudimentary horns are often associated with urinary tract anomalies, careful attention must be paid to the ipsilateral ureter and major uterine vessels during surgery. A study indicated that in non-communicating rudimentary horns, the ipsilateral ureter is located higher than usual because it lies close to the vascular connection of the rudimentary horn. Therefore, when transecting the round ligament and entering the broad ligament and retroperitoneal space, the ureter must be identified. Additionally, a firmly attached rudimentary horn may receive its blood supply not only from the ipsilateral uterine artery but also from myometrial arcuate arteries originating from the contralateral uterine artery, requiring meticulous hemostasis during dissection[17]. Although this case involved a communicating rudimentary horn, the pelvic anatomical variations remained relevant. Preoperative ultrasound indicated the patient had a solitary left kidney and right renal agenesis, obviating the need for identification of the right ureter during resection. However, we carefully dissected the adhesions and clearly delineated the vascular anatomy, noting that the internal and external iliac arteries ran parallel. Intraoperatively, we observed that the vascular branches supplying the rudimentary horn deviated significantly from normal uterine vasculature. After resecting the rudimentary uterine horn, we confirmed that both sacrouterine ligaments on the left side were intact, and the uterine arteries followed a normal course. However, the pelvic vasculature and nerve structures on the side of the rudimentary horn exhibited notable variations. Finally, we used laparoscopy and hysteroscopy to address the uterine fibroids in the unicornuate uterus. A transverse incision was made in the anterior uterine wall through the seromuscular layer to reach the pseudocapsule of a 4x4x3 cm fibroid, which was enucleated. Similarly, three fibroids were removed laparoscopically, followed by hysteroscopic resection of a 4x4x2 cm submucosal fibroid.

4. Conclusion

This case report presents a rare congenital uterine anomaly involving a rudimentary uterine horn, unicornuate uterus, and endometriosis. The patient underwent laparoscopic surgery to remove the rudimentary horn, ovarian cysts, and multiple uterine fibroids, with postoperative pathology confirming benign leiomyoma and adenomyosis. The case underscores the challenges in diagnosing rudimentary uterine horns due to non-specific symptoms and emphasizes the importance of combining clinical assessment with imaging for accurate diagnosis and treatment.

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