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(CASE REPORT)

A COMPLEX MÜLLERIAN ANOMALY MISDIAGNOSED AS FIBROID UTERUS: A CASE REPORT

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ABSTRACT

We report a 47-year-old woman who had never been pregnant and had suffered from primary infertility. She also had chronic pelvic pain and irregular menstrual periods. She had several uterine abnormalities, including a complete septate uterus, a duplicated cervix, and a longitudinal vaginal septum, with primary infertility. Her diagnosis included a large endometrial polyp and submucosal fibroids in the uterine cavity. This has been reported in only a few cases. The ultrasound diagnosis revealed a single uterine body with a single endometrial cavity and uterine fibroids but missed other diagnoses of two separate cervical canals, a double cervix, or separate vaginal canals, and a large endometrial polyp. The patient opted for definitive treatment and declined fertility preservation, leading to a total abdominal hysterectomy and removal of the uterine corpus, both cervices, and the vaginal septum. She was discharged five days after surgery and recovered. There are three clinical aspects to this case. First, imaging techniques (ultrasonography and MRI) should be used to diagnose and classify atypical Müllerian duct anomalies (MDAs) before applying a classification system. Second, in hysteroscopic septal resection, treatment should be individualized based on age, disease severity, and reproductive status. For younger women who wish to conceive, hysteroscopic septal resection is generally the most appropriate option, and hysterectomy is usually the last resort for suitable candidates. Finally, a multidisciplinary approach is necessary to deal with congenital anomalies that are complicated by acquired uterine pathologies. The diagnostic and surgical plan is required for such a patient to achieve optimal outcomes.

Keywords: Septate Uterus, Double Cervix, Müllerian Anomaly, Uterine Fibroid, Case Report

1 INTRODUCTION

Mullerian duct anomalies (MDAs) are congenital malformations caused by abnormal development, incomplete fusion, or failure of the paired mullerian ducts of the fetus. MDAs are found in the general population at 0.001% to 10% in women of reproductive age [1]. The septate uterus is found to be the most common MDA in women of reproductive age, about 2% to 3%, and is associated with the highest incidence of reproductive failure, such as infertility and recurrent pregnancy loss [2,3]. A fully septate uterus with cervical duplication and longitudinal vaginal septum is extremely rare [4].

Patients with this condition may have different symptoms like dysmenorrhea, dyspareunia, infertility, and recurrent miscarriage [1]. Moreover, acquired diseases, such as endometrial polyps and uterine fibroids, come up at the patient with this condition, too. This case report describes a 47-year-old woman with this type of MDA who was infertile, was also suffering from a large endometrial polyp and multiple fibroids, and required a total abdominal hysterectomy. This case illustrates the challenges of diagnosis, the need for high clinical suspicion, and the need for a multidisciplinary approach.

2 CASE PRESENTATION

A 47-year-old nulligravid woman came to the gynecology clinic with primary infertility and chronic pelvic pain. She had heavy menstrual bleeding for 10-12 days, frequently changing her pads and fatiguing herself, and was in pain. Her medical history was normal, and no surgical history or family history of congenital anomalies was known.

The general physical examination showed stable vital signs. Abdominal examination showed a non-tender, irregular, firm mass in the lower abdominal region, consistent with a gestational age of 14 weeks. Speculum examination showed a complete longitudinal vaginal septum from the introitus to the cervix, with two cervices on either side. Bimanual examination showed two cervices and an enlarged, irregularly shaped uterus. There was no tenderness on cervical motion or adnexal palpation.

Laboratory tests detected a hemoglobin level of 9.29/dL for which iron supplementation was given as required Table 1 [5].

Table 1: Laboratory reports showing anaemia

Parameters	Results	References value
Hb (g/dL)	9.2	10.65-15.35
RBC ($\times 10^{12}$/L**)**	3.32	3.81-5.5
WBC ($\times 10^{9}$/L)	5.6	2.85-8.46
Platelet ($\times 10^{9}$/L)	302.4	140.3-424.1
MCV (fL)	62.4	72.33-94.81
MCH (pg)	19.2	24.02-33.50
MCHC (g/dL)	26.6	32.94-36.76

Ultrasound showed a single uterus with several submucosal fibroids (the largest of which was 8.2×5.0×5.5 cm). There were no comments on a septate uterus, double cervix, double vaginal canal, or giant endometrial polyp, missing the complete diagnosis.

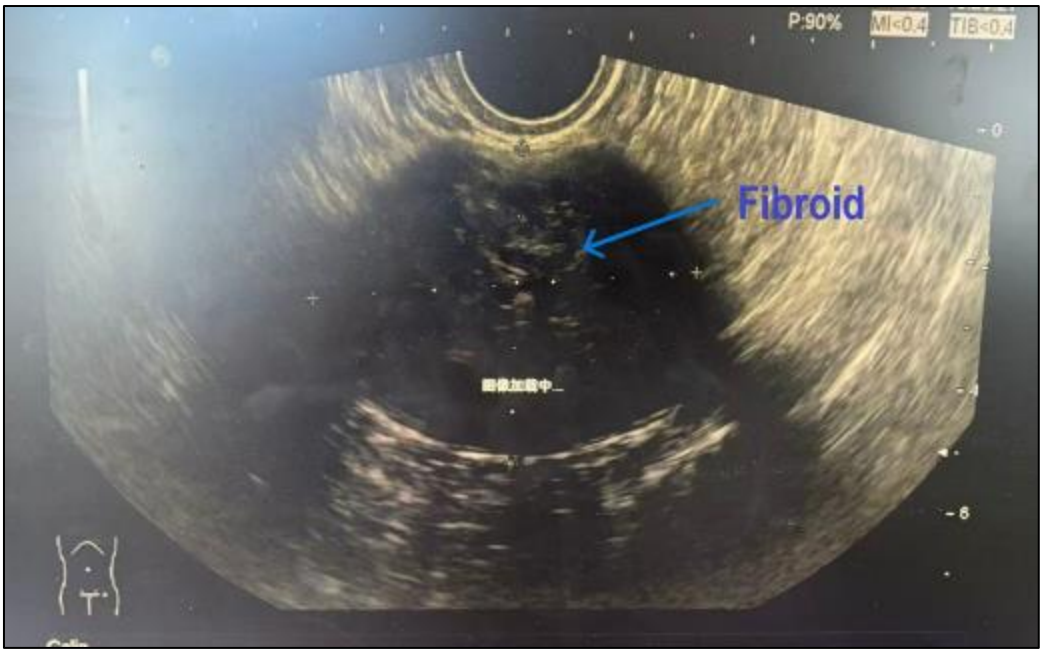


Figure 1: Ultrasound showing fibroid uterus

Considering the patient's age, nulligravid status, and multiple diseases, fertility-preservation options were not suitable. The patient had completed her reproductive years and had a desire for definitive management. In this case, total abdominal hysterectomy (TAH) was the most appropriate choice. The ovaries were preserved to maintain hormonal balance as she was premenopausal, and no adnexal pathology was observed. The patient was informed of the procedure and its risks and benefits, and she gave her consent.

The patient was put to bed under general anesthesia. A Pfannenstiel incision was made to access the abdomen; intraoperative results confirmed the radiological findings. A detailed survey of the pelvic cavity was conducted to determine and mobilize the ureters to prevent injury. The round ligaments, ovarian ligaments, and fallopian tubes were clamped, cut, and ligated bilaterally. The broad ligaments were incised, and the uterine arteries were clamped, cut, and ligated on both sides to avoid ureter and bladder injury. The remaining longitudinal vaginal septum was removed to the level of the introitus, and the single vaginal canal was formed. Hemostasis was achieved, and the vaginal cuff was closed with interrupted sutures. The pelvic peritoneum was reapproximated, and the abdominal incision was closed in layers after ensuring hemostasis.

The specimen was sent for histopathological examination. Grossly, an enlarged and distorted uterus with a thick fibrous septum dividing the endometrial cavity into two compartments, each with a cervix. Several fibroids were found beneath the myometrium, and a large endometrial polyp was identified in the right cavity after resection (Figure 2-5). Histopathology confirmed the uterine leiomyoma and endometrial polyp.

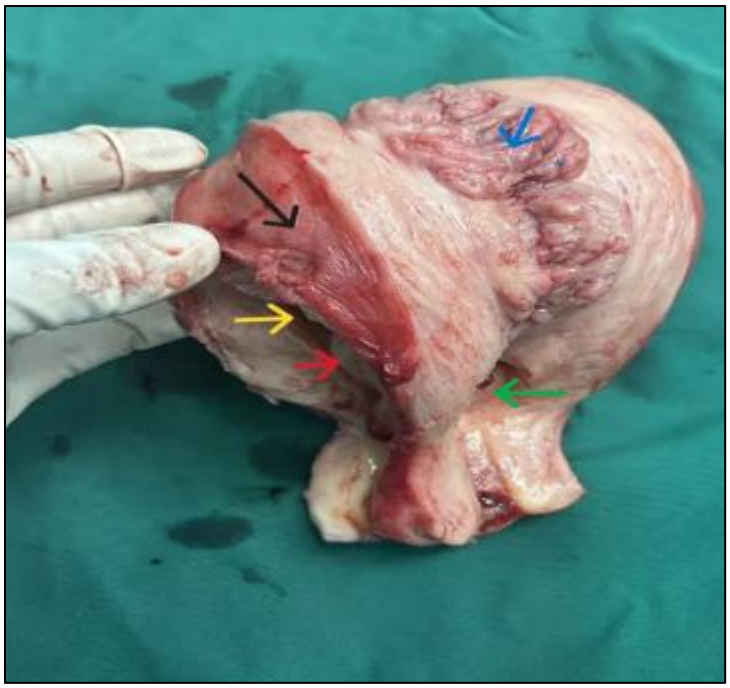


Figure 2: Image of resected uterus with labels indicated by arrows (red arrow: endometrial polyp; yellow arrow: right endometrial cavity; black arrow: thick uterine septum; green arrow: left endometrial cavity; blue arrow: fibroid).

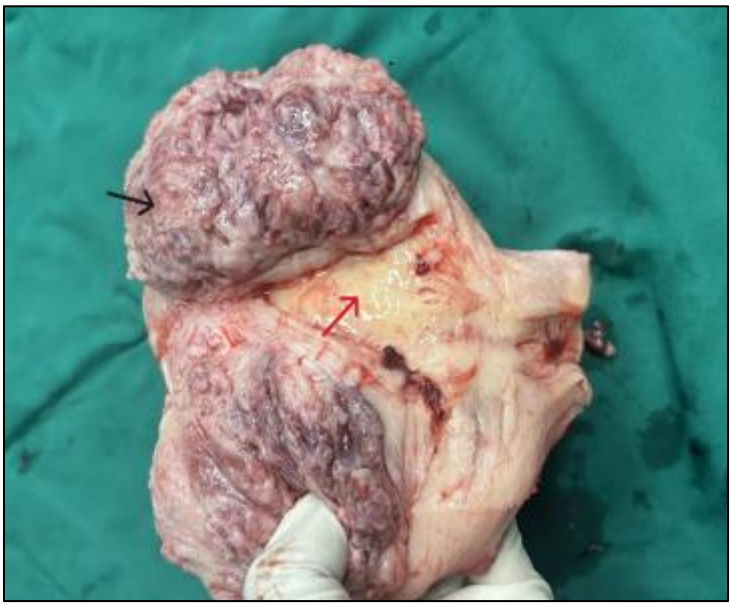


Figure 3: Image showing left endometrial cavity (red arrow), fibroid (black arrow)



Figure 4: Image showing right endometrial cavity (red arrow) and large endometrial polyp (black arrow)



Figure 5: Image showing two cervices indicated by black and red arrows.

The patient had a relatively uneventful recovery. She was ambulated on the first postoperative day and tolerated a regular diet. Pain management was done with oral analgesics, and hemoglobin levels were monitored with continued iron supplementation. She was discharged on the fifth day following surgery with wound care and follow-up appointments at 2 and 6 weeks. At the six-week follow-up, the patient was clear of bleeding and pelvic pain. A vaginal examination revealed a well-healed cuff and single vaginal canal, and she said she was pleased with the surgical outcome.

3 DISCUSSION

This case presents a rare combination of findings: a complete septate uterus with cervical duplication, longitudinal vaginal septum, infertility, endometrial polyp, and uterine fibroids. The rarity and complexity of this anomaly can lead to diagnostic delays or misdiagnoses [1]. Early suspicion could be due to examination of the vagina or cervix, where duplication may be observed. Diagnosis and classification are heavily dependent on the development of advanced imaging techniques [6]. MRI is one of the most reliable tools for the diagnosis of MDAs, but its accuracy for the specific

diagnosis of uterine septum may be as low as 70% in a few studies [7]. Three-dimensional transvaginal ultrasound (with or without saline infusion) is recommended as a first-line non-invasive diagnostic method since it has high accuracy (sometimes as high as 100% compared to operative diagnosis) [7].

The specific anomaly of a septate uterus, double cervix, and longitudinal vaginal septum is referred to as a Müllerian anomaly without classification. According to ASRM MAC 2021 classification, this anomaly is a variant of the septate uterus. Based on the ESHRE/ESGE classification, this anomaly can be classified as U2bC2V1 subtype [1]. The embryological reason for this is still unclear. The Müllerian duct fusion and resorption theory does not well define the presence of a septate uterus associated with a double cervix and vagina. This is consistent with the bidirectional Müllerian theory that the caudal and cranial pars of the Müllerian ducts developed independently [8].

Infertility is common for MDAs in women with a septate uterus [2]. It is difficult to estimate the true prevalence of infertility in this population because of selection bias. This affects fertility outcomes by making implantation less successful and leading to an increased chance of miscarriage [7]. In our case, infertility was compounded by the presence of an endometrial polyp and uterine fibroids. Endometrial polyps are common among women seeking fertility treatment and are thought to obstruct sperm transport and hinder embryo implantation [9]. Additionally, submucosal fibroids can distort the endometrial cavity, affecting endometrial receptivity and blood flow [10]. There is still research on surgical intervention for polyps and fibroids to improve infertility. Some observational studies have shown benefits, but high-quality randomized controlled trials are still lacking, particularly [11].

Based on her age and definitive treatment, the most appropriate approach for this patient was TAH. Different surgical options for the younger patients with fertility preservation may be available to them. Hysteroscopic resection of the septum is the standard surgical treatment for a septate uterus and is usually done laparoscopically for safety and to check the external uterine shape [12]. In cases of cervical duplication and vaginal septum, procedures like resection of the vaginal septum followed by hysteroscopic metroplasty can be performed [13]. In the case of a fibroid located on the septum, a two-step surgical procedure (myomectomy followed by hysteroscopic resection) may be used [14]. These disorders can be corrected by surgery, but the evidence is limited to case series [15].

4 CONCLUSION

This case study involves a 47-year-old woman presenting with a complete septate uterus, cervical duplication, and a longitudinal vaginal septum, further complicated by infertility, an endometrial polyp, and uterine fibroids. It highlights the diagnostic and clinical complexities associated with rare Müllerian duct anomalies. Accurate diagnosis requires a high degree of suspicion, complemented by effective imaging modalities such as ultrasound and MRI. Management strategies should be individualized, taking into account factors such as the patient's age, symptoms, and reproductive goals. For patients experiencing infertility, surgical intervention to restore normal anatomy and address concurrent pathologies should be considered. In this case, a total abdominal hysterectomy was deemed necessary, illustrating that surgical intervention can be definitive in certain scenarios. Future research, particularly larger prospective studies, is crucial for understanding the treatment of these rare and complex anomalies and establishing clear, evidence-based guidelines.

STATEMENTS ON COMPLIANCE WITH ETHICAL STANDARDS

Disclosure of conflict of interest

No conflict of interest to be disclosed.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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