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Case Report

Spontaneous intrauterine pregnancy, post laparoscopic surgery for didelphys uterus with longitudinal vaginal septum, multiple leiomyomas and bilateral proximal tubal block

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ABSTRACT

Mullerian duct anomalies (MDAs) are congenital developmental anomalies of the female reproductive tract. Uterine didelphys also known as a double uterus, with two uteri and cervices, and a single or double vagina, is one of the rarest MDA. Uterine leiomyomas are a common finding in the normal uterus but extremely rare in women with MDAs. We present the case and surgical management of a 28 year-old African woman with uterus didelphys with two separate cervix and vagina with multiple leiomyomas and bilateral proximal tubal block. She presented with inability to conceive and dyspareunia and was referred to us with a diagnosis of multiple fibroids uterus. She was managed successfully by a combined hysteroscopic, laparoscopic and vaginal approach with myomectomy and excision of longitudinal vaginal septum. Post procedure she conceived spontaneously after 6 months. Management of this case was a challenge owing to the coexistence of multiple complex malformations and the difficulty in preserving the reproductive potential. The aim of this case report is to expand the available knowledge about this group of females affected with a common diagnosis of leiomyomas, superimposed on an extremely rare MDA, uterine didelphys.

Keywords: Congenital mullerian anomalies, Didelphys uterus, Leiomyoma, Longitudinal vaginal septum

INTRODUCTION

Congenital uterine anomalies exist in approximately 4.3% of fertile women.[1] Any deviation in the dynamic process of differentiation, before 22 weeks of gestation, can result in a wide spectrum of complex uterine anomalies with cervical, vaginal and renal malformations. Uterine didelphys is a type of rare congenital uterovaginal anomaly accounting for 5-11% of all MDAs.[2] Renal abnormalities may be seen in conjunction with non-obstructed uterine didelphys in 15-30% of population.[3] A complete longitudinal vaginal septum is seen in 75% of women with obstructive uterine anomalies.[4] Uterine abnormalities, both congenital and acquired are significant causes of infertility and obstetric complications. Patients with a didelphys uterus are usually asymptomatic, but some present late with dyspareunia, dysmenorrhea, hematocolpos and infertility. We present the case of a 28 years old African female who presented with inability to conceive and dyspareunia, which was misdiagnosed as multiple fibroids uterus in her country. On evaluation she was found to have uterus didelphys with two separate cervix and vagina with multiple leiomyomas and bilateral proximal tubal block. Treatment involved surgical resection of the longitudinal vaginal septum with hysteroscopy guided bilateral fallopian tube catheterization and laparoscopic myomectomy. Fibroids, though rare in the context of MDAs can complicate diagnosis, treatment, and reproductive outcomes. Surgical management depends on several factors, including the patient's desire for future fertility and the leiomyoma type and position.

CASE REPORT

A 28 years old African female was referred to us with complaints of dyspareunia and inability to conceive for 5 years. She was diagnosed as multiple fibroids uterus on transvaginal ultrasound examination in her country and was offered a surgical treatment. Her menstrual cycles were regular. Her gynecological examination revealed normal external genitalia with a complete longitudinal vaginal septum with two separate cervix. Magnetic resonance imaging (MRI) revealed uterus didelphys with two separate uterus with endometrial cavities with two cervix and vagina with multiple leiomyomas, largest being 4×2 cm in right uterus and 1.9×1 cm in left uterus. Bilateral ovaries and adnexa were normal. Kidneys and pelvicalyceal system showed no apparent abnormality. Infertility workup was within normal limits. Husband semen analysis was within normal limits. On the basis of clinical examination and radiologic studies, diagnosis of didelphys uterus with longitudinal vaginal septum with multiple leiomyomas was made. After detailed counseling and informed consent, decision of hysteroscopy and laparoscopic myomectomy with excision of longitudinal vaginal septum was taken.

Under general anesthesia abdomen was entered by 1 cm supraumbilical primary port. Pneumoperitoneum was created and ipsilateral ports placed. Diagnosis of didelphys uterus was confirmed at laparoscopy. Each side uterus was seen with ipsilateral tube and ovary attached to them. Two fibroids of size 2×2 cm and 3×3 cm fibroids were seen in right uterus and one 1×1 cm fibroid was noted in left uterus (Figure 1).

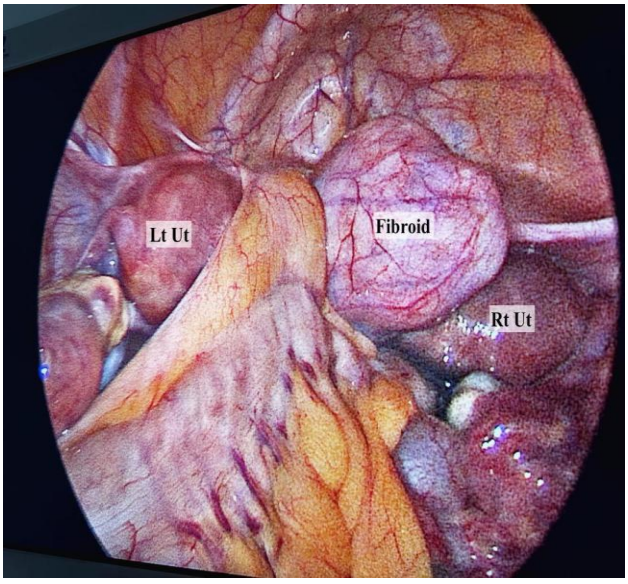

Figure 1: Laparoscopic view suggesting uterus didelphys with fibroids. Right uterus (Rt Ut), Left uterus (Lt Ut), Fibroid.

On vaginal examination longitudinal vaginal septum was noted till introitus (Figure 2).

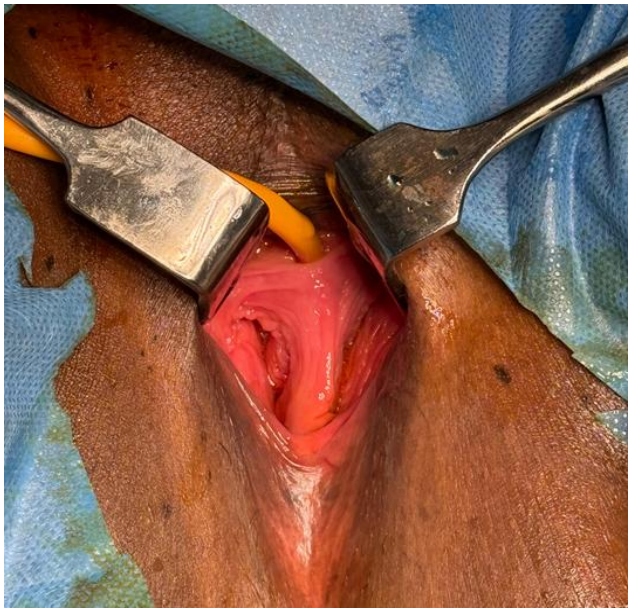

Figure 2: Perineal view showing a longitudinal vaginal septum extending till introitus.

Two separate cervix and patent endocervical canal were noted. The operative hysteroscope was then inserted gently through the cervix, and the endometrial cavities were observed separately in right and left uterus. Chromopertubation test (CPT) done with indigo blue dye, was negative on both sides. Bilateral proximal tubal occlusion was noted. Bilateral fallopian tube catheterization (FTC) was done with no. 3 French FTC catheter. Repeat CPT was positive for both fallopian tubes. Fibroids were demarcated and vasopressin in dilution of 1 in 100 ml normal saline was injected into the stalk. Enucleation and resection of fibroids was done using electrocautery. After resection of the leiomyoma the myometrium was closed in two layers with V-Lok barbed sutures. After securing hemostasis intercede was placed on suture site to prevent adhesions.

Lastly longitudinal vaginal septum resection was done and raw area approximated with 2-0 vicryl absorbable suture. Hemostasis was confirmed at the end of the procedure with an estimated blood loss of 300 ml and operative time of 2 hours and 30 minutes. Myoma was retrieved from supraumbilical port and send for histopathological examination (HPE).

Postoperative she was discharged in a stable condition after 2 days with instructions to follow up in one week. Regular vaginal dilatation was advised with plastic dilators to maintain patency. HPE report suggested benign leiomyoma. Two months after the surgery, the patient did not have any problems during sexual intercourse. Patient was instructed not to plan pregnancy for 6 months. She did not come for any further follow-up visits. Post procedure she spontaneously conceived after 6 months and ultrasound showed an early intrauterine pregnancy of 5 + 4 weeks (Figure 3).

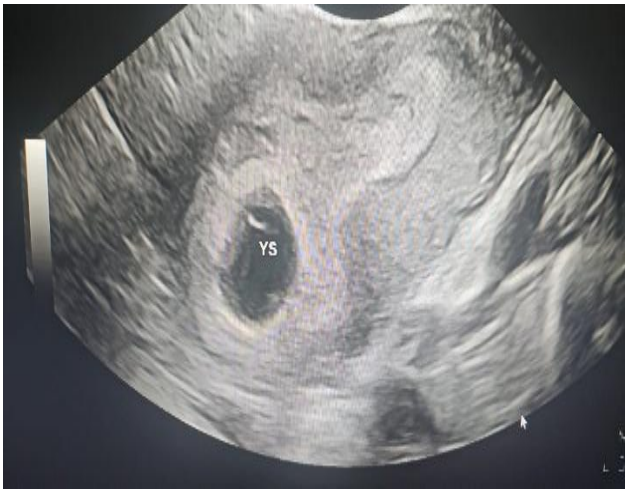

Figure 3: Ultrasound image suggesting an early intrauterine pregnancy.

*yolk sac (YS).

DISCUSSION

Uterus, cervix, fallopian tubes, and upper two-thirds of the vagina originate from the mullerian ducts, whereas the lower vagina is formed from the urogenital sinus.[5] Complete lack of fusion of the mullerian ducts between 12th and 16th weeks of gestation, results in uterine didelphys with two widely divergent non communicating uteri, two cervices, associated sometimes with a vaginal septum.[6] MDAs are usually associated with age-appropriate external genitalia and functioning ovaries.[4]

Approximately 40% of cases are associated with renal malformations owing to the close embryological development of the mesonephric and mullerian ducts during the ninth week of gestation.[7] Lateral fusion defect of the vagina from caudal to cranial direction can result in a vaginal septum.[8] Herlyn-Werner-Wunderlich-Syndrome was described in the 1980's which included triad of didelphys uterus, ipsilateral renal agenesis and obstructed hemi-vagina.[9] To allow inclusion of other uterine anomalies, the acronym OHVIRA (obstructed hemivagina and ipsilateral renal agenesis) was devised in 2007.[9] While didelphys uterus may occur in isolation, it is sometimes seen associated with bladder exstrophy, congenital vesicovaginal fistula, cervical agenesis, double urethra, double sacrum, double ureters, multiple vertebral anomalies, cloacal anomaly and anorectal malformation.[4,6]

Classification system for congenital uterine anomalies was proposed by Buttram Jr. and Gibbons in 1979, and it was revised by the American Fertility Society in 1988.[10] In 2021, the American Society of Reproductive Medicine expanded it to include a wider range of mullerian anomalies.[1] Uterine leiomyomas are one of the most common benign gynecologic tumors in women of reproductive age with an estimated presence of 77%.[1] The International Federation of Gynecology and Obstetrics classification (FIGO) classifies uterine fibroids from 0 to 8 based on their relationship to the uterine muscle wall and cavity. It helps in identifying and planning the optimal surgical approach for myomectomy.[1]

The first imaging modality for diagnosis of MDAs is abdominopelvic ultrasound, but the MRI is a key imaging modality because it can classify the malformation and identify concomitant anomalies.[7] Other diagnostic modalities include invasive methods like hysteroscopy, hysterosalpingography (HSG), and laparoscopy/ laparotomy. In selected cases laparoscopy can be of great help for diagnosis.[7] Giannella et al reported a misleading case of fibroid in a woman with a unicornuate uterus which was found to be nonfunctional rudimentary horn on diagnostic workup. This study enhances the need for preoperative imaging modalities in cases of suspicious mullerian anomalies.[11]

Majority of patients are asymptomatic until menarche, but eventually present as chronic abdominal pain, dysmenorrhea, dyspareunia, palpable abdominal mass, hematocolpos, hematometra, endometriosis, pelvic inflammatory diseases and fertility issues.[7] The association between major uterine fusion defects and fertility outcomes is debatable. Review by Grimbizis et al found that the incidence of MDAs in infertile patients (3.4%) is similar to that of the fertile women (4.3%).[10] Zeleke et al found comparable outcomes in women with a didelphys uterus and other types of MDAs but lesser than with a normal uterus.[2]

Study by Crowley et al in women with MDAs showed increased risk of adverse obstetric outcomes.[9] Pregnancies complicated by MDAs showed increased risks of miscarriage, ectopic pregnancy, cervical incompetence, preterm labour, fetal growth restriction, fetal malpresentation, labor dystocia and caesarean section.[2,9] Ludmir et al in their study found that with high-risk obstetric intervention available, fetal survival rate was higher in didelphys uterus in comparison to the bicornuate and septate group.[10] In patients with OHVIRA syndrome, successful pregnancies have been reported in up to 87% of cases with 62% patients delivering at term without complications.[7] In patients with history of obstetrical complications and fertility issues, surgical treatment appears to be the best option.

Treatment options are usually tailored to the specific mullerian anomaly and targeted to improve the quality of life and increase fertility.[4] Metroplasty or surgical unification of a didelphys uterus is not usually indicated. Apparent benefits of metroplasty are not clear and most results are anecdotal.[4] Surgical correction of longitudinal vaginal septum in the non-obstructed didelphys uterus is reserved for patients with severe dyspareunia.[4] In patients with uterus didelphys with obstructed unilateral vagina, the longitudinal vaginal septum is excised in its entirety to relieve the obstruction and visualize both cervices.[12] Attaran et al demonstrated pregnancy rates of 87% and live birth rates of 77% post septum excision.[12] Excision of the

vaginal septum and laparoscopy for associated fibroids, endometriosis, adhesions or infertility can be performed as a single procedure to decrease long-term morbidity and preserve reproductive function.[4] Caliskan et al report laparotomy and myomectomy surgery, for a leiomyoma on the septum of a septate uterus, followed by in vitro fertilization and conception in view of bilateral tubal block.[13] Hemihysterectomy with or without salpingo-oophorectomy should be avoided unless complicated by persistent endometriosis, adenomyosis, or when other surgical treatments have failed.[4] Studies show that in didelphys uterus, vaginal delivery should be considered first unless indications for cesarean delivery are present.[10] Routine cerclage is not indicated in didelphys uterus unless there is a history of cervical incompetence or premature births.[10]

CONCLUSION

Due to low incidence and lack of awareness about didelphys uterus, it is often diagnosed late or misdiagnosed. When a didelphys uterus is diagnosed, searching for any concurrent renal and genital anomalies is useful. Interpretation of ultrasound and MRI can be challenging in mullerian anomaly with fibroids, but they play a vital role in surgical decision making. Surgical correction of didelphys uterus is not usually indicated, however if the women is symptomatic, excision of the obstructing vaginal septum may be required. Presence of leiomyomas, poses challenge due to radiologic difficulty of distinguishing the deviation from normal anatomy. As demonstrated by our case report timely and accurate recognition of didelphys uterus with multiple fibroids and early surgical resection of the vaginal septum was crucial in maintenance of fertility and good obstetric outcome.

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