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Website: www.pogsjournal.org

DOI: 10.4103/pjog.pjog_53_24

Case report on the management of ectopic pregnancy in uterine didelphys

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Abstract:

The case report discusses a rare occurrence of tubal pregnancy in a patient with uterine didelphys, managed using combined hysteroscopy and laparoscopy. A 29-year-old Gravida 2 Para 0 (0010) with a right tubal ectopic pregnancy alongside incidental uterine didelphys detected via physical examination and three-dimensional transvaginal ultrasound. The patient underwent a combined diagnostic and operative laparoscopy, where a right salpingectomy was performed using a harmonic scalpel. Diagnostic laparoscopy showed two uterine horns with each attached fallopian tube and ovary with an interostial length of 4 cm. Diagnostic hysteroscopy confirmed the presence of two separate uterine cavities and cervices without communication. The procedure demonstrated that the technique for laparoscopic salpingectomy in cases of uterine didelphys parallels that for a normal uterus. The use of combined hysteroscopy and laparoscopy proved effective in evaluating both the external uterine structure and internal cavity, facilitating accurate diagnosis and treatment of Müllerian anomalies with ectopic pregnancy.

Keywords:

Hysteroscopy, laparoscopy, tubal pregnancy, uterine didelphys

Introduction

An ectopic pregnancy occurs when a fertilized egg implants outside the uterus, commonly in the fallopian tube's ampullary region, affecting 1%–2% of women. Risk factors include age, smoking, and prior ectopic pregnancies.[1,2] Uterine didelphys, a rare Müllerian anomaly, affects 6.3% of women and is associated with kidney abnormalities.[3] It carries a 2.3% incidence of ectopic pregnancy, with higher rates of miscarriage (20.9%) and preterm delivery (24.4%), but most women (68.6%) achieve live births.[4] This report details a case managed via combined hysteroscopy and laparoscopy, crucial for both diagnosing the anomaly and treating the ectopic pregnancy.

Case Report

A 29-year-old woman, Gravida 2 Para 0 (0010), presented with a history of vaginal spotting for 2 weeks and amenorrhea for 5 weeks. Increased vaginal bleeding and hypogastric pain led her to seek emergency care at a tertiary hospital. She had a previous abortion in 2019. She had regular menses lasting 3–4 days with moderate flow. She denied dyspareunia, sexually transmitted infections, and contraceptive use, having had coitarche at 19 and two sexual partners. Physical examination revealed a soft abdomen. On speculum examination, the cervix had two external oses, and there was brownish vaginal discharge. On internal examination, the cervix was firm and movable with no tenderness or adnexal masses. The initial impression was threatened abortion, which cannot rule out ectopic pregnancy, to consider Müllerian anomaly. Her serum beta-human chorionic gonadotropin (B-hCG) was 1326 mIU/mL. Transvaginal ultrasound with two-dimensional (2D) and 3D imaging showed a right adnexal mass measuring 4.09 cm × 3.05 cm × 2.05 cm with a volume of 16.38 cm^3, superior to the right ovary. The uterus had two converging uterine horns at

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Submitted: 30-Jul-2024
Revised: 02-Aug-2024
Accepted: 20-Aug-2024
Published: 26-Dec-2024

How to cite this article: Morales AR, Cagalawan RM, Alcantara-boquiren MJ. Case report on the management of ectopic pregnancy in uterine didelphys. Philipp J Obstet Gynecol 2024;48:279-82.

the level of the internal cervical os, with two cervices. The external fundal indentation completely divided the uterine corpus up to the cervix. The left kidney was seen and the right kidney was not visualized. The preoperative diagnosis was Tubal Ectopic Pregnancy Right, Unruptured; Müllerian Anomaly Uterine Didelphys with Right Unilateral Renal Agenesis.

The patient underwent operative laparoscopy with right salpingectomy and a diagnostic hysteroscopy under general anesthesia. On laparoscopy, there was no hemoperitoneum noted. There were two uterine horns noted each with an attached fallopian tube and an ovary with an interostial length of 4 cm [Figure 1]. The right fallopian tube was dilated to 5 cm × 3 cm × 5 cm with products of conception inside [Figure 2]. The liver and subdiaphragmatic areas were smooth and without adhesions. A right salpingectomy was done. The fimbrial end of the right fallopian tube was grasped with atraumatic forceps. A series of cutting and coagulation of the mesosalpinx from the uterotubal junction toward the fimbrial end was done using harmonic scalpel. Serial coagulation and cutting of the raw edges were performed using Bipolar Maryland Forceps on 40W energy. A diagnostic hysteroscopy using a 30-degree rigid hysteroscope revealed two cervical openings with a midline indentation and no connection between them [Figure 3]. Inside, two banana-shaped uteri were observed, each with a visible ostium, and no communication between them was noted [Figures 4 and 5]. The patient tolerated the surgery well without complications and was discharged after 2 days. She was advised to do hysterosalpingography 6 weeks postsurgery. The final diagnosis was Gravida 2 Para 0 (0020) Tubal Pregnancy Ampullary, Right Unruptured; Müllerian Anomaly Uterine Didelphys, Unilateral Renal Agenesis.

Discussion

Müllerian anomalies are congenital defects of the female genital system that occur from abnormal embryological development of the Müllerian ducts.[3] These defects can be brought about by the failure of differentiation, migration, fusion, and canalization of the Müllerian duct system which usually happens during the 6th and 22nd week in utero.[3,5]

Uterine didelphys was found to be the second least common Müllerian anomaly with a prevalence of 8.3%.[6] Most women with uterine didelphys are usually asymptomatic. However, some women may present with dyspareunia, dysmenorrhea, or experience difficulty in tampon insertion, associated with a longitudinal vaginal septum.[3] Patients with Müllerian anomalies, such as unicornuate and didelphic uteri, may also exhibit renal anomalies. This is attributed to the developmental relationship between Wolffian ducts and Müllerian ducts, where kidney abnormalities can coincide with

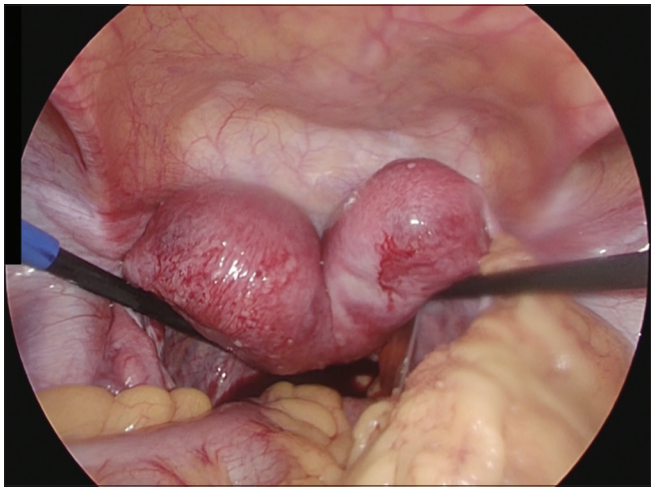

Figure 1: Two hemi-uterus with fallopian tubes

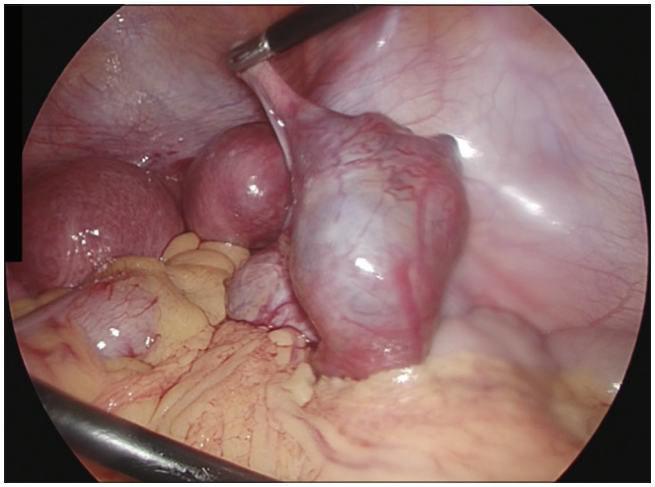

Figure 2: Right tubal pregnancy

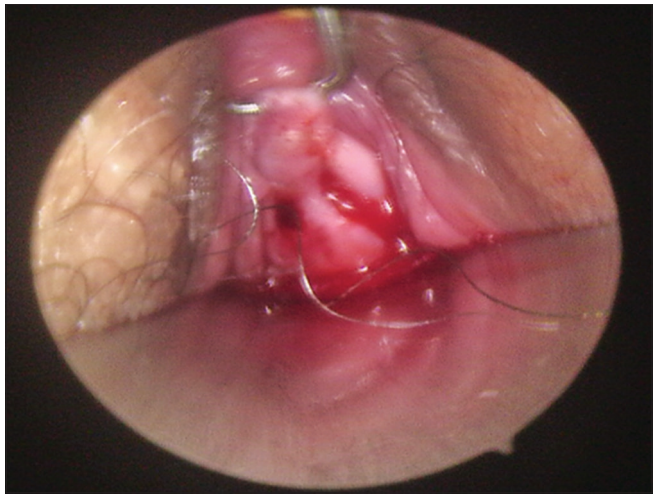

Figure 3: Hysteroscopy: 2 cervical openings

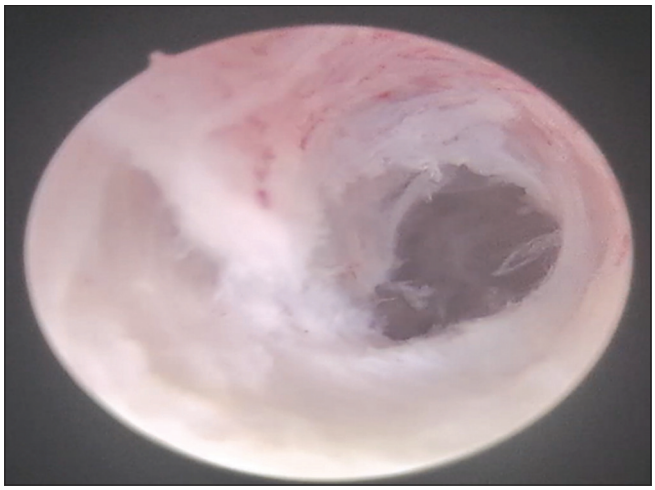

Figure 4: Left hemi-uterus with adhesion and ostium

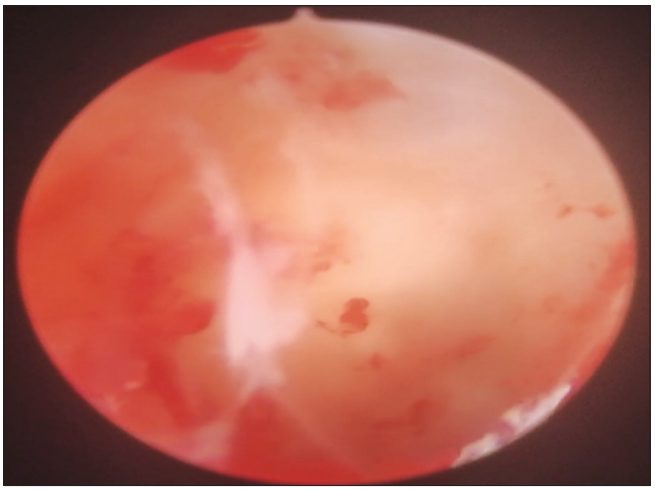

Figure 5: Hysteroscopy, right hemi-uterus

uterine abnormalities.[7] As it was evident in this case, there was unilateral renal agenesis as seen on ultrasound.

The use of 2D transvaginal ultrasound and hysterosalpingogram in diagnosing Müllerian anomalies is less invasive and can be used for screening patients. However, its use is limited since these modalities cannot accurately classify the type of Müllerian anomaly. The use of 3D transvaginal ultrasound and magnetic resonance imaging (MRI) offers better diagnostic accuracy since they define both internal and external uterine contours. In the past, combined laparoscopy and hysteroscopy were considered the best method for diagnosing Müllerian anomalies. However, recent studies indicate that 3D ultrasound has surpassed them as the new gold standard. This shift is due to 3D ultrasound's superior diagnostic accuracy, less invasive procedure, and greater cost-effectiveness.[4] In this case, the use of a combined hysteroscopy and laparoscopy evaluated the external uterine contour and excluded other uterine malformations. The diagnostic hysteroscopy evaluated the uterine cavity and ostia, the absence of connection between the two hemi-uteri, and the presence of two cervices, establishing the diagnosis of uterine didelphys. The pregnancy outcomes associated with this type are similar to unicornuate uterus with a term delivery rate of 45%.[6]

The incidence of an ectopic pregnancy occurring in a woman with a didelphys uterus is 2.3%.[4] In this case, the patient presented with vaginal spotting and hypogastric pain with a positive pregnancy test. On 2D transvaginal ultrasound, showed an extrauterine pregnancy at the right adnexal area, two uterine horns, and two cervices. 3D transvaginal ultrasound also showed the external fundal indentation that completely divided the two uterine corpora up to the level of the cervix.

Ectopic pregnancy has an overall incidence rate of 1%–2% in the general population and 2%–5% incidence rate among women who have undergone assisted reproductive technology.[1] The recurrence rate of ectopic pregnancy in a patient with one history of ectopic pregnancy is 10% while in two or more prior ectopic pregnancies, the rate is 25%.[8] Diagnosis of ectopic pregnancy includes serum B-hCG measurement in correlation with transvaginal or transabdominal ultrasound.[8] The discriminatory zone of B-hCG 1000–2000 mIU/mL in diagnosing ectopic pregnancy has been established. It is expected to appreciate a gestational sac confirming an intrauterine pregnancy by transvaginal ultrasound above the level of the discriminatory zone.[1] In this case, the patient had a serum B-hCG of 1326 mIU/mL. On transvaginal ultrasound, both the right and left uterus had thickened endometria and there was no evidence of an intrauterine gestational sac. Instead, a heterogeneous mass located superolateral to the right ovary with circumferential vascularity was seen.

Management of ectopic pregnancy may either be medical or surgical. Indications for medical management with single-dose methotrexate can be done in asymptomatic and compliant patients with <3.5 cm size of ectopic gestation, without fetal cardiac activity, and BHCG values <5000 mIU/mL. In the case presented, the patient underwent laparoscopic salpingectomy of the right fallopian tube. Since the size of the ectopic gestation was more than 3.5 cm and the patient was already symptomatic with vaginal bleeding and hypogastric pain, she was not a good candidate for medical management. Three prospective randomized trials showed the superiority of doing a laparoscopic approach over laparotomy including less blood loss, less pain medication requirement, shorter length of hospital stay, and faster postoperative recovery.[1] Salpingectomy is preferred in cases where there is rupture and

uncontrolled bleeding, tubal damage, prior tubal sterilization, or a tubal mass >5 cm in diameter.[1] Laparoscopic right salpingectomy was done on the tubal ectopic pregnancy, similar to that of a normal uterus. The technique entails removing the tube from its anatomic attachment using bipolar energy, ultrasonic energy, or end loops. The incision starts at the uterotubal junction progressing to the mesosalpinx until reaching the fimbrial ends.

The incidence of recurrent ectopic pregnancy is approximately 15%, which rises to approximately 30% after 2 previous episodes of ectopic pregnancies.[1] The assessment of tubal patency of the contralateral fallopian tube can be done by hysterosalpingography after 6–8 weeks postsurgery to allow the inflammation to subside and improve spontaneous pregnancy outcome.[9]

Conclusion

Tubal ectopic pregnancy in uterine didelphys is rare due to its lowest prevalence among congenital uterine anomalies. Diagnosis typically involves 3D transvaginal ultrasound or MRI. Hysterolaparoscopy is used to assess Müllerian anomalies by examining both external and internal uterine structures. Salpingectomy procedures are comparable in patients with uterine didelphys and those with a normal uterus. Further studies can be done to determine reproductive and pregnancy outcomes of patients with uterine didelphys.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Authorship contributions

Arriane R. Morales, MD - Involved in the conceptualization, methodology, data curation, writing of the original draft, review and editing

Ricca Mae G. Cagalawan, MD - Involved in the conception, design, provision of resources, journals for review of literature, patient care, and follow- up.

Marie Janice Alcantara-Boquiren, MD - Involved in the conception methodology, supervision, review and editing of the draft.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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