Content available at: https://www.ipinnovative.com/open-access-journals

Indian Journal of Obstetrics and Gynecology Research

Journal homepage: www.ijogr.org

Case Report

Postmenopausal bleeding with a diadelphic uterus: A case report

Aishwarya Choudhari[1,*], Senthil Kumar A C[2], Kundavi Shankar[3]

[1]*Institute of Reproductive Medicine and Women's Health, Madras Medical Mission, Chennai, Tamil Nadu, India*
[2]*Madras Medical Mission, Chennai, Tamil Nadu, India*
[3]*Dept. of Reproductive Medicine and Women's Health, Madras Medical Mission, Chennai, Tamil Nadu, India*

ARTICLE INFO

Article history:
Received 23-05-2023
Accepted 24-06-2023
Available online 24-08-2023

Keywords:
Diadelphic uterus
Postmenopausal bleeding
Mullerian anomalies
Altered anatomy

ABSTRACT

Background: Simple uterine anomalies to more complicated multisystem derangements can all fall under the broad category of congenital abnormalities known as mullerian duct anomalies. Urological and caudal gastrointestinal abnormalities may be linked to complete uterine, cervix, and vaginal duplication.
Purpose: This case report aims to raise awareness of the challenges surgeons face while treating Mullerian abnormalities and to foresee ureter and artery supply duplication.
Case Report: In this case report, 59 year old, P1L1, Previous LSCS with diadelphic uterus with longitudinal vaginal septum presented with postmenopausal bleeding. MRI of the patient showed endometrial hyperplasia in both uteri with no obvious myometrial infiltration. Patient was planned for hysterectomy frozen section and proceed SOS to staging laparotomy in view of recurrent postmenopausal bleeding and morbid obesity. Total abdominal hysterectomy with bilateral salphingophorectomy with frozen section done. During surgery bladder was adherent to previous c section scar on the left horn, left sub total hysterectomy was done and sent for frozen followed by right total hysterectomy. Along with the right total hysterectomy sample both cervices were delivered. Frozen section showed a simple hyperplasia without atypia in left horn and hyperplasia with foci of intraepithelial neoplasia in right horn, Final histopathology further confirmed the same report.
Conclusion: It's important to have a full awareness of any potential related deformities when managing patients with Mullerian defects and irregular uterine haemorrhage. To prevent urological problems and surgical mishaps, a thorough preoperative evaluation, meticulous surgical investigation, and multidisciplinary approach may be required.

1. Introduction

Congenital flaws of the female genital system known as Mullerian duct abnormalities result from the improper embryological development of the Mullerian ducts. One of these abnormalities can be the absence of development, fusion, canalization, or reabsorption, which commonly occur between 6 and 22 weeks in utero. The majority of publications place the prevalence of these anomalies in the general population between 0.5 and 5.0%.[1–4]

The septate uterus, followed by the bicornuate uterus and the arcuate uterus, has a mean incidence of about 35% and is the most common uterine defect.[4] According to general consensus,[1–5] uterine anomalies are linked to worse pregnancy outcomes, including higher probabilities of spontaneous abortion, early labour, caesarean delivery because of breech presentation, and fewer live births. The severity of these effects differs among various uterine abnormalities, though.

* *Corresponding author*.
E-mail address: aishchoudhari1@gmail.com (A. Choudhari).

https://doi.org/10.18231/j.ijogr.2023.071

Four major categories are obvious when categorising these anomalies merely based on aberrant development.

Complete or partial failure of Mullerian duct formation; Failure of ducts to canalise (unicornuate uterus with a primitive horn lacking suitable chambers); Agenesis; unicornuate uterus lacking even the most basic horn. Mullerian ducts that have not fully fused (bicornuate or didelphys uterus)[6,7]uterine septum (septate or arcuate uterus) incomplete reabsorption.[4] A didelphys uterus has two cervices and separate uterine compartments because the Mullerian ducts have completely failed to join. A longitudinal vaginal septum is also present; it can be either thin and elastic or thick and inelastic. A routine speculum exam usually yields the first indication of the problem, which is then followed by a diagnosis, when anatomical anomalies are seen and call for additional examination. Additionally, because the Mullerian and Wolffian ducts frequently co-develop, it is possible to find kidney abnormalities alongside uterine abnormalities.[1,2] Many Mullerian anomalies are also associated with renal anomalies and need through diagnosis before opting for any surgery.

2. Case Report

Patient is a postmenopausal woman, age 59. Paragraph 1 Living 1. In order to evaluate postmenopausal bleeding in the context of a thicker endometrial stripe, the patient visited a gynaecology clinic. She was known to have a diadelphic uterus, which is a congenital abnormality.

Her menarche occurred at the age of 12, and she had a history of one caesarean delivery following a full-term pregnancy in the left uterus. She had been nine years out of menopause. She had her final period when she was 50 years old. She had complains of recurrent postmenopausal bleeding for which she was being evaluated. Patient had pipelle biopsy in 2020 and HPE- showed disordered proliferative endometrium. She had never used hormone replacement therapy before. She was up to date on pap smear screening and had no prior history of abnormal pap smear results. Patient had a family history of breast cancer, and underwent FNAC twice for a breast lump- which had findings of benign fibroadenosis. She has been having annually checkups in breast clinic.

Up until five months before to the appointment, when she experienced a five-day episode of intermittent vaginal bleeding from both vagina, she was in generally good condition. Labia that were widely apart and duplicated vagina and cervix were among the physical examination findings.

Her initial gynaecological evaluation included transabdominal ultrasound testing, which revealed a larger endometrial stripe in her right hemiuterus (10 mm) and a normal endometrial stripe in her left hemiuterus (6 mm). MRI abdomen and pelvis was planned and showed uterine diadelphys (Figure 1) with endometrial hyperplasia with no obvious evidence of myometrial. Bilateral kidneys noticed and no urological malformations noted on MRI (Figure 3).

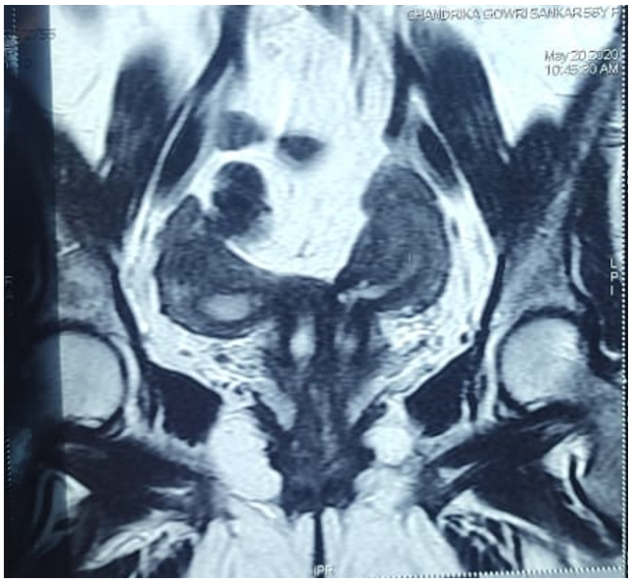

Fig. 1: Diadelphic uterus with two horns, two cervices and two vaginas

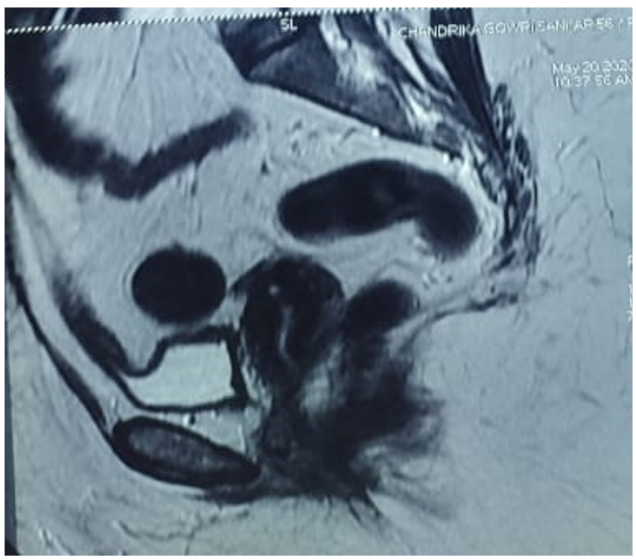

Fig. 2: Sagittal section

Her endometrial thickness and BMI were taken into account, and an oncologist's opinion was sought regarding the possibility of endometrial cancer. After undergoing considerable counselling, the patient chose to move forward with the removal of both uteri and the frozen section examination with likely staging depending on these findings. She also decided, regardless of the outcome of the

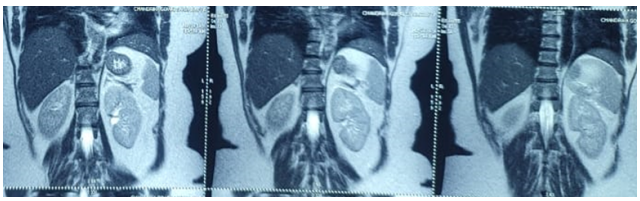

Fig. 3: Bilateral kidneys seen without any anomaly

frozen section, to have a bilateral salpingo-oophorectomy at the time of the scheduled surgery.

The procedure involved a midline incision. Following entry into the abdominal cavity, a pelvic examination revealed two uteri that were widely spaced apart, each with its own fallopian tube and ovary, for a total of two fallopian tubes and two ovaries (Figure 4). As previously explained, each uterus and cervix communicated with its own distinct vagina. There were no obvious visual abnormalities in the pelvis or abdomen, and no free fluid was visible. The bilateral cervices are widely covered by the UV fold. Retroperitoneal space was bilaterally invaded prior to hysterectomies. A tracing of the ureters on both sides revealed no anatomical abnormalities.

After first clamp of infundibulopelvic ligaments cutting and securing, bladder was tried to pushed down. Bladder was adherent to left uterus over the previous caesarean section scar, hence subtotal hysterectomy done of the left uterus (Figure 5) and send for frozen section. Right uterus total hysterectomy performed and during this both the cervices (Figure 6) were delivered and sample sent for frozen section. Frozen section reported as proliferative endometrium in the left uterus and hyperplasia with foci of intraepithelial neoplasia in the right uterus. Post hysterectomy vault was closed and later bladder integrity tested by instilling methylene blue dye. Both the bladder and hemostasis were intact. The surgery was finished at this moment because the frozen part did not show any malignancy. 200ml of blood was reportedly lost during the treatment. Bilateral salphingo-oophorectomy and total abdominal right and left hysterectomy were the final procedures.

3. Discussion

In this example, a patient with postmenopausal haemorrhage and a thicker endometrial stripe was given a typical clinical scenario that called for additional testing to rule out an endometrial precancerous disease. Despite the fact that the final pathological evaluation did not find any evidence of cancer, the care and evaluation stages were adequate. A thorough understanding of the embryologic causes of these anomalies and the potential for associated malformations is necessary when planning the surgery.

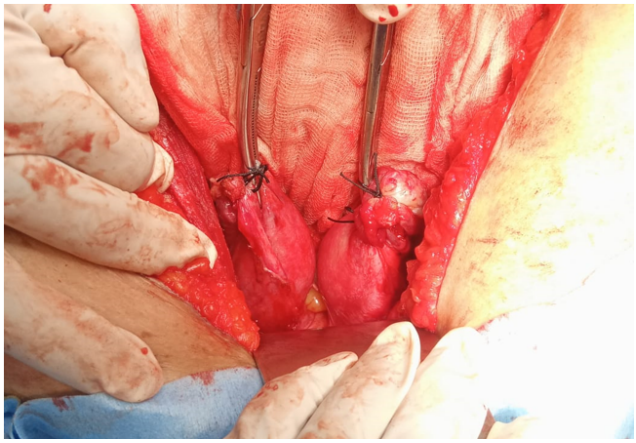

Fig. 4: Diadelphic uterus with bilateral adnexa

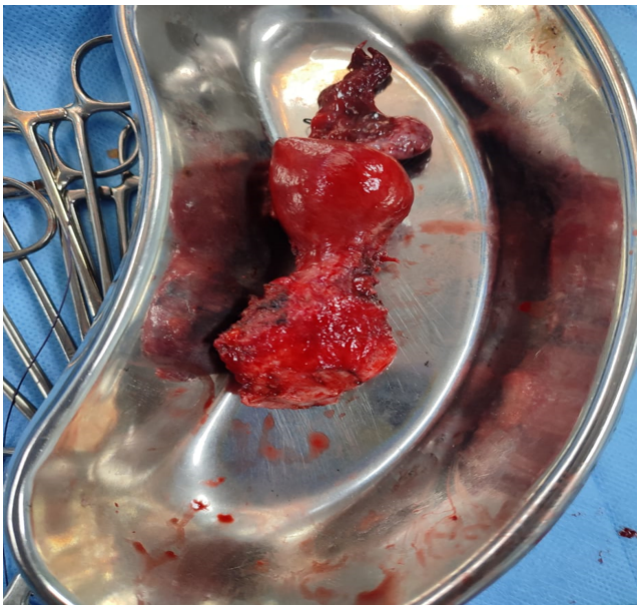

Fig. 5: Left subtotal hysterectomy

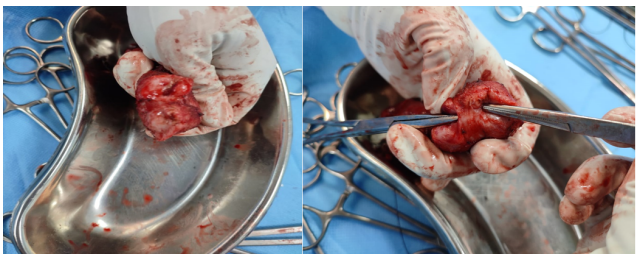

Fig. 6: Bilateral cervices delivered along the right total hysterectomy specimen

Comparatively speaking to other Mullerian duct anomalies listed in the ASRM 2021 classification,[8] a didelphys uterus is still a relatively uncommon defect. Didelphic uteri have been demonstrated to be less than those with normal uterine anomalies but better than those with other Mullerian duct abnormalities. The majority of this anomaly's 11% uterine abnormalities are asymptomatic.

Based on the type of defect and the fault in embryologic development, the precise anatomy varies. The nephrogenic cord and the gonadal ridge, which later give rise to the urinary and genital systems, respectively, are developed on both sides of the dorsal aorta by the fourth week of embryonic development. Both male and female embryos have two pairs of genital ducts by the fifth and sixth weeks: mesonephric (wolffian) and paramesonephric (mullerian). The paramesonephric ducts develop even in the absence of male developmental cues.

The uterovaginal canal is created by the fusion of the mullerian ducts, which begins caudally and moves cranially. The non-fused sections of the mullerian ducts develop into the fallopian tubes. The development of the Mullerian ducts in a female embryo is thought to be significantly influenced by the female gubernaculum, an embryonic organ. It is thought to have a muscular origin and, by connecting to the mullerian ducts, it promotes or perhaps forces the uterus and fallopian tubes to develop normally. As the female gubernaculum continues to mature, it continues to exist as the circular ligament that penetrates the abdominal wall and the ovarian ligament.[9] Urinary tract and lower GI systems develop nearby in space and time to the reproductive system.

A blind-ended caudal hindgut develops during the fourth week of development. The cloaca is the hindgut's most distal end, and by the seventh or eighth week, it is divided into the anorectal canal and the urogenital sinus, which later give rise to the urinary bladder and urethra.[10] When the latter makes contact with the uterovaginal primordium, sinovaginal bulbs—paired outgrowths—are produced. The centre portion of the vaginal plate, which eventually fuses to form the sinovaginal bulbs, breaks down and creates the lower side of the vaginal lumen.[11]

Female genital anomalies have been categorised using the American Fertility Society (AFS) classification system, the embryological-clinical classification system, and the Vagina, Cervix, Uterus, Adnexa, and associated Malformations (VCUAM) classification system, with the AFS system being the most well-liked.[12] According to the AFS system, the 7 distinct groups of female genital anomalies include:

1. Segmental or complete agenesis or hypoplasia
2. Unicornuate uterus with or without a rudimentary horn
3. Didelphys uterus
4. Complete or partial bicornuate uterus
5. Complete or partial septate uterus
6. Arcuate uterus, and
7. Diethylstilbestrol (DES) - related anomalies.

The groups I and II, III and IV, and V and VI, from an embryological perspective, represent a stoppage in one of the following processes: early development, fusion, or degeneration. Sadly, this classification does not describe additional related abnormalities.[13] In order to correctly identify each aberration, ESHRE created a taxonomy based mostly on uterine anatomy, with cervical and vaginal malformations classified as separate supplemental subclasses.[14] The ASRM Mullerian Anomalies Classification 2021 (MAC2021) divides mullerian anomalies into nine categories: mullerian agenesis, Cervical agenesis, Unicornuate uterus, Uterus didelphys, Bicornuate uterus, Septate uterus, Longitudinal vaginal septum, Transverse vaginal septum, and Complex anomalies.[8] MAC 2021 also describes the variants in these anomalies and helps in diagnosis and appropriate treatment.

Postmenopausal bleeding could indicate a number of diseases, such as uterine cancer, atrophic endometrium, endometrial polyps, and endometrial hyperplasia. 10% of individuals with postmenopausal bleeding have endometrial cancer, which calls for a complete work-up that involves tissue collection and sonographic examination of the pelvic organs. Not unexpectedly, this patient's complicated anatomy made managing her post-menopausal haemorrhage difficult. She was already evaluated for recurrent postmenopausal bleeding and had undergone endometrial biopsy previously showing endometrial hyperplasia. She was scheduled for a staging laparotomy, however due to changing anatomy, retroperitoneal dissection and ureter identification were required. Because the bladder was attached to the scar from the c-section, careful dissection was done during the left hysterectomy in order to protect the ureter and the vasculature of the pelvic side wall.

4. Conclusion

An in-depth knowledge of potential related abnormalities is necessary for the management of individuals with Mullerian defects and irregular uterine bleeding. To prevent urological problems and surgical mishaps, a multidisciplinary approach, cautious surgical investigation, and thorough preoperative examination may be required.

5. Source of Funding

None.

6. Conflict of Interest

None.

References

1. Heinonen PK. Uterus didelphys: a report of 26 cases. *Eur J Obstet Gynecol Reprod Biol*. 1984;17(5):345–50.

2. Heinonen PK. Clinical implications of the didelphic uterus: long-term follow-up of 49 cases. *Eur J Obstet Gynecol Reprod Biol*. 2000;91(2):183–90.
3. Raga F, Bauset C, Remohi J, Bonilla-Musoles F, Simón C, Pellicer A. Reproductive impact of congenital Müllerian anomalies. *Hum Reprod*. 1997;12(10):2277–81.
4. Grimbizis GF, Camus M, Tarlatzis BC, Bontis JN, Devroey P. Clinical implications of uterine malformations and hysteroscopic treatment results. *Hum Reprod Update*. 2001;7(2):161–74.
5. Acién P. Reproductive performance of women with uterine malformations. *Hum Reprod*. 1993;8(1):122–6.
6. When things go wrong: congenital anomalies. South Africa: University of Cape Town; 2011. Available from: https://health.uct.ac.za/pathology-learning-centre/disease-themes/when-things-go-wrong-congenital-anomalies.
7. Chandler TM, Machan LS, Cooperberg PL, Harris AC, Chang SD. Müllerian duct anomalies: from diagnosis to intervention. *Br J Radiol*. 2009;82(984):1034–42.
8. Pfeifer SM, Attaran M, Goldstein J, Lindheim SR, Petrozza JC, Rackow BW, et al. ASRM müllerian anomalies classification 2021. *Fertil Steril*. 2021;116(5):1238–52.
9. Acién P, delCampo FS, Mayol MJ, Acién M. The female gubernaculum: role in the embryology and development of the genital tract and in the possible genesis of malformations. *Eur J Obstet Gynecol Reprod Biol*. 2011;159(2):426–32.
10. Schoenwolf GC, Larsen WJ. Larsen's human embryology. 4th ed. Philadelphia: Churchill Livingstone/Elsevier; 2009.
11. Moore KL, Persaud TVN, Torchia MG. The developing human: Clinically oriented embryology. 10th ed. Philadelphia, PA: Elsevier; 2016.
12. Sardo ADS, Campo R, Gordts S, Spinelli M, Cosimato C, Tanos V, et al. The comprehensiveness of the ESHRE/ESGE classification of female genital tract congenital anomalies: A systematic review of cases not classified by the AFS system. *Hum Reprod*. 2015;30(5):1046–58.
13. Robbins JB, Broadwell C, Chow LC, Parry JP, Sadowski EA. Mullerian duct anomalies: Embryological development, classification, and MRI assessment. *J Magn Reson Imaging*. 2015;41(1):1–12.
14. Grimbizis GF, Gordts S, Sardo ADS, Brucker S, DeAngelis C, Gergolet M, et al. The ESHRE/ESGE consensus on the classification of female genital tract congenital anomalies. *Hum Reprod*. 2013;28(8):2032–44.

Author biography

Aishwarya Choudhari, Fellow in Reproductive Medicine

Senthil Kumar A C, Surgical Oncologist

Kundavi Shankar, HOD

Cite this article: Choudhari A, Kumar A C S, Shankar K. Postmenopausal bleeding with a diadelphic uterus: A case report. *Indian J Obstet Gynecol Res* 2023;10(3):366-370.