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Uterus Didelphys: A Case Series

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Abstract
Uterus Didelphys is a rarest congenital anomaly of Mullerian ducts. It is mostly asymptomatic but found to be associated with breech presentation, preterm births, miscarriages and cesarian sections. This anomaly may be associated with longitudinal vaginal septum leading to dyspareunia and inability to conceive and obstruction during labor. Here, I report a series of four cases of uterus didelphys all with complete longitudinal vaginal septum with conception after resection of vaginal septum in one of them; while three conceived without any problem with septum. Three of these were term pregnancies while one was preterm with history of recurrent miscarriages. All were breech presentations and delivered by cesarian section.

Keywords: Uterus didelphys, longitudinal vaginal septum, mullerian ducts, breech presentation, cesarian section

Introduction
Mullerian anomalies are developmental disorders of female reproductive tract resulting in various uterine, cervical and vaginal malformations. They are common conditions with prevalence of 5.5% in general population and 24.5% in patients treated for miscarriage and infertility [1]. Most common amongst these are septate uterus and bicornuate uterus approximately 35% and 25% respectively [2]. Uterus didelphys is rarest amongst them and accounts for 10% of Mullerian anomalies [3]. Uterus didelphys occurs due to arrest of fusion of Mullerian ducts which can be complete and incomplete resulting in duplication of uterine horns and cervix [4]. Duplication of vagina can be either complete (75%) or incomplete [5]. Renal agenesis is found to be associated in 20% cases [5]. According to ASRM Mullerian anomalies classification 2021 it is classified as Uterus didelphys and+/- longitudinal vaginalseptum of variable length [6].
Most women with this disorder are asymptomatic; while some of them may present with dyspareunia and/or dysmenorrhea [7]. Though uterus didelphys rarely affect fertility it often leads to obstetric complications such as recurrent miscarriages, preterm births, malpresentations, intrauterine growth restriction and low birth weight babies.While most of the cases remain undiagnosed until evaluated for other gynecological disorders [7,8]; cases with fetal malpresentations diagnosed during pregnancy or at the time of delivery are one of the commonest causes of Caesarian delivery in these cases [9]. Here I present a case series of four cases of uterine didelphys with clinical presentations ranging from difficulty in conception, recurrent miscarriages and spontaneous conception with breech presentation to preterm deliveries.

Case Presentations
Case 1: A 24year old women presented to Obstetrics and Gynecology OPD with complain of inability to conceive with married life of 4years; on further detailed history; she also complained of dyspareunia. She attained menarche at age of 13 years; menstrual cycles were regular with history of spasmodic dysmenorrhea. Her general examination and systemic examination was normal, Per Abdomen was soft and non-tender. On P/S examination a complete longitudinal vaginal septum was noted extending from introitus up to cervix, also two external cervical os were noted on either sides of the vaginal septum. On P/V examination: the longitudinal vaginal septum was felt approximately 7-8 cm in length and 1-1.5cm in thickness extending up to cervix; two cervical os were felt.
Uterus could not be felt in midline by bimanual examination.
Patient and her husband were counselled regarding the findings. Upon investigating her routine

investigations, thyroid and prolactin levels were normal; semen analysis showed normospermia; on Trans Vaginal Sonography Mullerian anomaly was suspected which was found to be Didelphys uterus on MRI. No renal anomaly was found.

Diagnosis was confirmed by Hystero-laparoscopy which showed two separate uterine cavities; each connected with fallopian tubes; two ovaries were seen which normal were looking. Complete longitudinal vaginal septum was removed surgically and two cervical os were clearly seen. Upon hysteroscopy two normal cervical canals; two uterine cavities which were small and equal in size with normal looking endometrium noted. On Chromopertubation test of each uterine cavity separately; both fallopian tubes were found to be patent.

Following the treatment patient conceived naturally within a year. At 16 weeks pregnancy was noted in left uterine cavity on USG and cervical length was normal. Routine antenatal follow up was done. At 38 weeks after discussion with patient and her husband; elective LSCS was done for breech presentation and average size baby. A single alive male baby was delivered of 2.75 kg. baby had cleft lip and palate. Patient's postoperative period was uneventful and she was discharged in good condition.

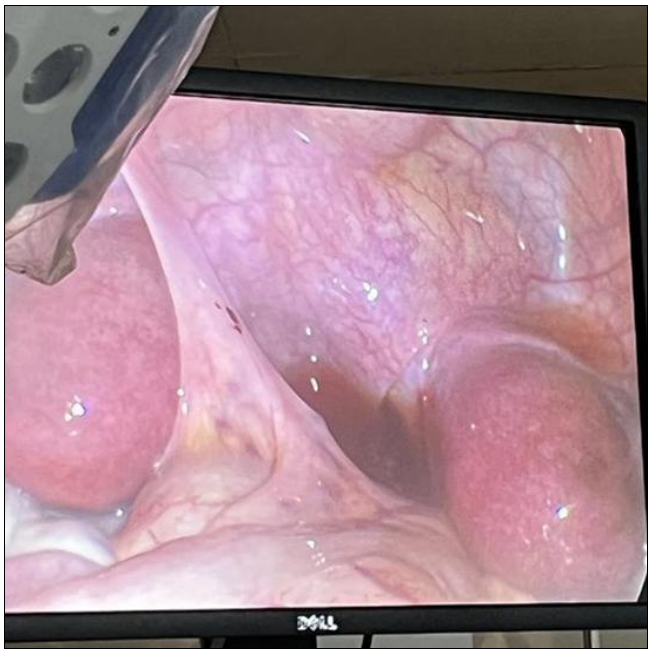

Fig 1: Two separate uterine fundi seen laparoscopically

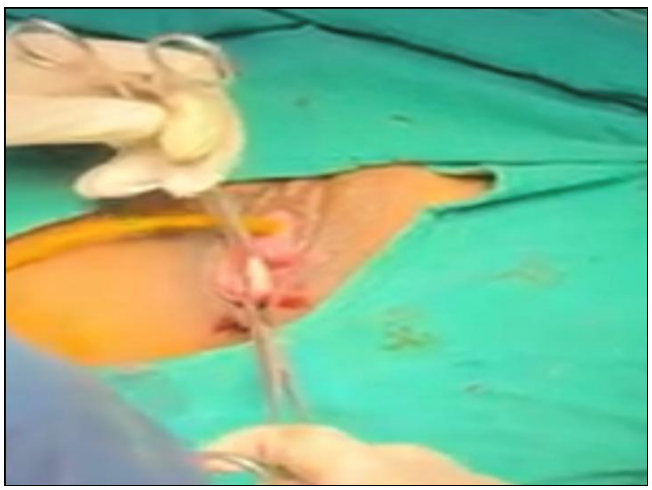

Fig 2: Vaginal septum as seen at introitus

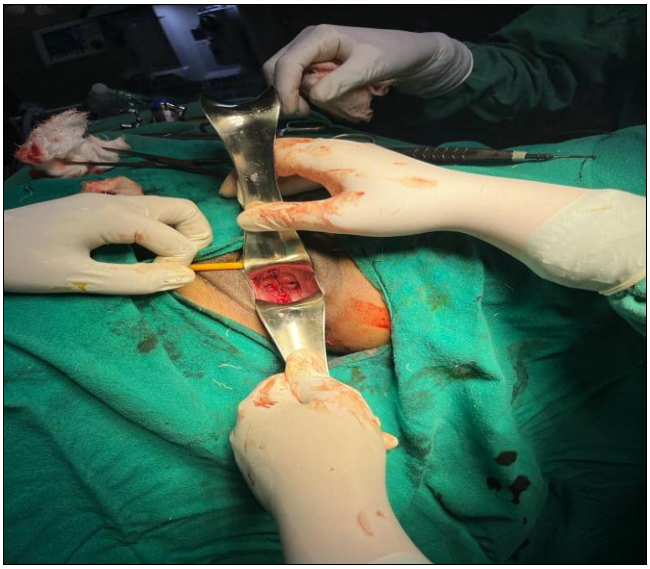

Fig 3: Two cervical os as visualized after removal of vaginalseptum

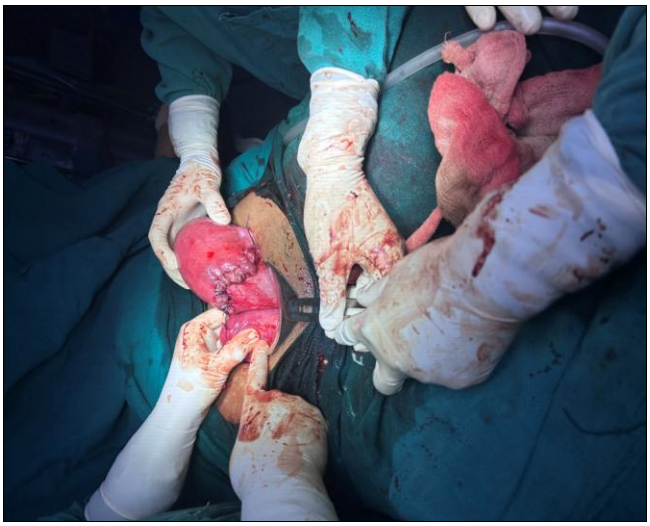

Fig 4: Stitched left uterine horn after delivery of the baby by LSCS along with right uterine horn

Case 2: A 21 year old primigravida at 38weeks and 6 day POG presented in emergency with complaint of pain abdomen for 6hrs; not associated with bleeding or leaking pervaginum and there was no history of decrease fetal movements. She was married for 1year and concieved spontaneously. Her antenatal period was uneventful and there was no significant past history. She attained menarche at age of 14 yrs and menstrual cycles were regular associated with mild spasmodic dysmenorrhea. There was no history of dyspareunia. Her past medical records were found to be normal.

Her general examination was normal; vitals were normal. On per abdomen examination fundal height was term size, longitudinal lie, breech presentation, mild contractions were present and FHS was 145 bpm and regular. On per vaginum examination a longitudinal vaginal septum was felt approx.8 cm long and 1.5 cm thick and two cervical os were felt with left cervical os 1.5 cm dilated and 20% effaced while right cervical os was closed. Patient and her attendants were counselled and an emergency LSCS was done for breech presentation and thick vaginal septum; with delivery of single alive female child of 3 kg. Per operatively Uterus was found to be didelphic with fetus in left uterine horn. Each uterine cavity is found to be connected to fallopian tube which were normal and both ovaries were also

normal. Patient and her husband were unaware of the condition till then. Her postnatal period was uneventful. On Post operative day 4 her USG abdomen was done and no renal anamoly was found. Patient was discharged in good condition.

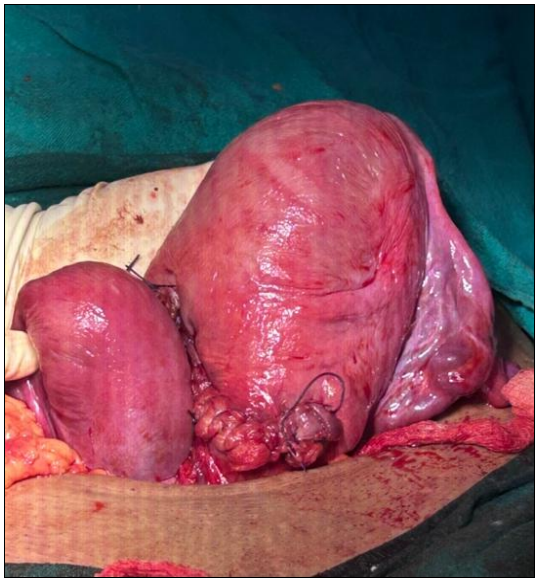

Fig 5: Stitched left uterine horn after delivery of the baby by LSCS and right uterine horn seen separately; both uterine horns are connected to right and left fallopian tubes

Case 3: A 22-year-old primigravida patient presented in emergency at 37 weeks and 4 days POG with complaints of pain abdomen for 8 hrs. with no history of bleeding per vaginum, leaking per vaginum and decrease fetal movements. She attained menarche at age of 13 yrs.; with regular menstrual cycles and not associated with dysmenorrhea. She was married for 2years. There was no history of dyspareunia and no significant past history. General and Systemic examination was within normal limits. On Per abdomen examination Fundal height was 36 weeks, longitudinal lie, breech presentation, FHS was 140 bpm with mild uterine contractions present. On P/V examination a longitudinal complete vaginal septum was felt approx. 8x1.5cm and two cervical os were also felt with right os found to be 3cm dilated and 30% effaced with absent membranes with footling presentation; station was -3 and left os was closed and uneffaced. Emergency LSCS was done for footling breech presentation and two well-developed uterine horns were noted with pregnancy in right horn and a single alive female baby weighing 2.5 kg was delivered by breech extraction. Each uterine cavity is found to be connected to fallopian tube which were normal and both ovaries were also normal. Patient and her husband were unaware of the condition till then. No renal anomaly was found on USG done in post operative period and patient was discharged on POD 5.

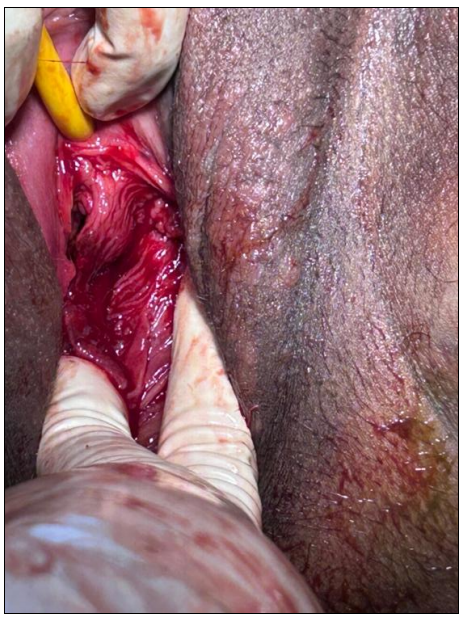

Fig 6: Longitudinal vaginal septum at the introitus

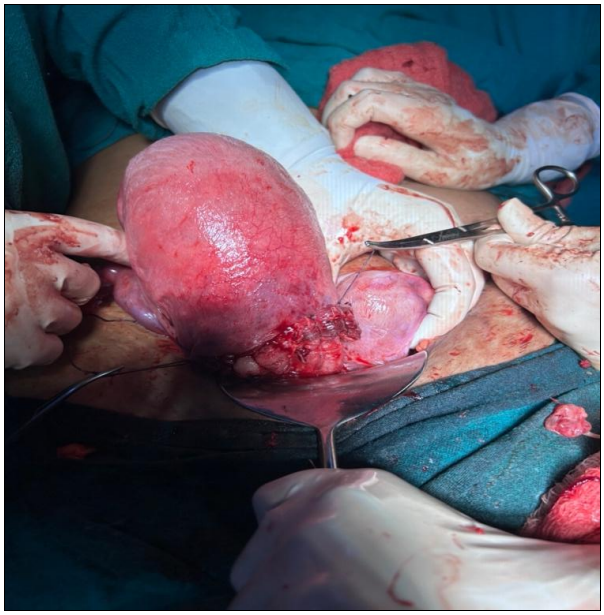

Fig 7: Stitched larger right uterine horn after delivery of the baby by LSCS along with smaller left uterine horn

Case 4: A 25-year-old women G2P0+0+2+0 married for 3 years presented in emergency at 35 3/7weeks gestational age with complain of leaking per vaginum for 5 hrs. There was no history of pain abdomen, bleeding per vaginum or decrease fetal movements. She had history previous2 spontaneous miscarriages at 3 and 3.5 months of amenorrhea 1year and 2 years back respectively. She attained menarche at 13 years of age; had regular menstrual cycles associated with mild spasmodic dysmenorrhea. There is no history of dyspareunia. On Per Abdominal examination fundal height was 36 weeks; fetus was in longitudinal lie with breech presentation FHS was 115 bpm, with relaxed uterus. On P/S examination a thick longitudinal complete vaginal septum was present and 2 cervical os were noted on either side of the septum.

Leaking was present from right cervical os and liquor was clear. On P/V examination right cervical os was found to be 1.5cm dilated and 10% effaced with absent membranes with station at -3; whereas left cervical was closed and uneffaced.

NST was found to be non-reassuring and patient was taken for emergency LSCS. During emergency LSCS two separate uterine cavities were found with fetus in right uterine cavity; and a single alive female baby was delivered by breech extraction and birth weight was found to be 2.35 kg on further examination each uterine cavity is found to be connected to fallopian tube which were normal and both ovaries were also normal. Patient and her husband were unaware of the condition till then. Patient had uneventful post-operative period and discharged on POD 5. No renal anomaly was found on USG.

Discussion

Uterus didelphys is a rare congenital anomaly of female reproductive tract accounting for 8% of all anomalies [2]. It is found in 0.3% of total population while 2.1% in females with history of miscarriages and infertility [1]. This anomaly is mostly asymptomatic; however, it can present with dysmenorrhea; dyspareunia; hematocolpos and hematometra [7]. It can besuspected with 2D USG and Hysterosalpingography however; accurate diagnosis can be made with 3D USG and MRI [10].

Congenital uterine anomalies have an impact on reproductive health of women and uterine didelphys is one amongst them. Being mostly asymptomatic; the diagnosis is usually delayed and it is detected first time when patient present in labor as in my three of the cases. It also doesn't impair ability to conceive but significantly leads to obstetrical complications. In a report of 26 cases by Heinonen; dysmenorrhoea, dyspareunia and leukorrhoea were most common symptoms [11]. Resection of vaginal septum has been found to relieve dyspareunia [12].

Similarly in my first case patient had dyspareunia which was relieved after longitudinal vaginal septal resection and she conceived naturally; while other three patients didn't have problem in conceiving.

A metanalysis by Venetis *et al* of congenital uterine anomalies showed uterus didelphys was not associated with decreased natural or assisted fertility; but significantly associated with spontaneous abortions, preterm births, breechpresentation, intrauterine fetal growth restriction; low birth weight and perinatal death [13]. In all the above four cases patients has breech presentation; and one of the patients had history of recurrent miscarriages.

In long term follow up of 49 women with didelphic uterus Heinonen P.K. has reported longitudinal vaginal septum was excised in 53% and 18% had obstructed hemivagina [14]. In my study all four patients had complete longitudinal vaginal septum; and one it was resected to relieve dyspareunia. Although vaginal deliveries have been reported in women with uterus didelphys; Cesarian section rate was found to be high as high as 84% [14]. Vaginal septum has been reported to cause dystocia [14] can also lead to obstruction of labor. In my study all four patients had breech presentation three of them being term pregnancy and one with preterm pregnancy. All of them delivered by cesarian section one due to footling presentation other for non-reassuring NST and other due to patients concerns about risk associated with abnormal uterus and vaginal septum.

Three out of four cases have been diagnosed during cesarian section one being diagnosed prenatally. There have been case reports of uterus didelphys not detected in previous one or even two caesarian sections there by increasing the risk of life-threatening uterine rupture in next pregnancy [15,16]. Thus, emphasizing the complete observation of uterus and adnexal structures during each cesarian section.

Conclusion: Uterus didelphys poses challenges to obstetrician and gynecologist in terms of diagnosis due to absent or minimal

symptoms and signs. High index of suspicion with complete physical examination and investigation is required. Dyspareunia due to vaginal septum leading to difficulty to conceive can be corrected by septal resection. Pregnancies with uterus didelphys are considered to be high risk due to associated obstetrical complications. In cases of Uterus didelphys with thick vaginal septum and associated patient's concerns cesarian section should be considered as a joint decision of medical team and patient.

Conflict of Interest
Not available.

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