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Case Report

A Case Series of Uterine Didelphys Discovered Incidentally During Child Birth

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Abstract

Uterine didelphys is the rarest form of uterine anomaly. It occurs as a result of failure of or abnormal fusion of the paramesonephric duct. It is mostly asymptomatic but may occationally present with some Gynaecological or Obstetrics complications. Due to it rarity, it is often overlooked during evaluation of patient that present with some of its symptoms. In most cases it is discovered incidentally as it is seen in these 2-case series and other cases in literature. Case A was an incidental finding of uterine didelphys, longitudinal vaginal septum and a right renal diverticulum in a woman with 2 previous caesarean section discovered at the 3rd caesarean section. Case B was that of acute inversion of one uterus in a primigravida with uterine didelphys masquerading as an endometrial polyp discovered during polypectomy. She had subtotal hysterectomy of the inverted extra uterus. The management of uterine didelphys is often individualized. Factors such as type of anomaly, symptomatology, associated complications and patient specific need often determine treatment offered.

Keywords: Didelphys, Uterine, Anomaly

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Introduction

Uterine anomaly can go unrecognized especially where there are no gynaecological or obstetric reasons that may prompt evaluation and recognition. This often occur due to rarity of the condition and low index of suspicion in asymptomatic patients. Müllerian duct anomalies (MDA) are a spectrum of congenital defects arising from the failure of fusion, abnormal formation or resorption of the mullerian duct during organogenesis.[1] Failure of the mullerian duct fusion results in bicornuate or didelphys uterus.[2] Uterine didelphys or double uterus result from complete failure of fusion of ipsilateral mullerian duct. This result in formation of a hemi-uteri each with its separate ovary and fallopian tube, cervix and often 2 separate vaginas in about 75% of cases.[3] The prevalence of mullerian abnormality ranges from 0.5-6.7% in general population.[4-6] Uterine didelphis is the rarest of all the other forms of mullerian anomalies. It accounts for about 10% of mullerian anomalies. In majority of cases, its often asymptomatic during childhood and puberty accounting for the delay in diagnosis until during pregnancy.[5, 7] However, it may present with dysmenorrhea, dyspareunea and infertility. While during pregnancy, it may present with miscarriage, premature rupture of fetal membranes (PROM), intra uterine growth restriction, preterm labour, malpresentation, renal agenesis, post-partum haemorrhage, caesarean section and reduced chances of life birth.[5] Diagnosis is often incidental as reported in most case report.[8] In suspected cases, diagnosis can be made with the aid of a transvaginal sonography, sono-hysterography, hysterosalpingography, hysteron-laparoscopy and pelvic magnetic resonant imaging.[7, 9] The American society for reproductive medicine (ASRM 1988) classifies uterine didelphys as class 3, while a more comprehensive classification by the European society for human reproduction and embryology (ESRE 2013) classifies it as U3b/C2.[10, 11]

We present a case series of 2 incidental finding of uterine didelphys in Katsina state.

Case A

A 28yr old booked $G_3P_2^{+0}A_2$, with 2 previous caesarean section at 38 weeks and 1 day, with breech presentation. She was admitted for repeat elective caesarean section. She had no labour pains or drainage of liquor and she perceived fetal movement. All her booking parameters were normal. Except persistent breech presentation, which was the case in both her last pregnancies. Her gynaecological history was unremarkable. She had no history of dysmenorrhea or dyspareunia She had no known medical condition. She had 2 sisters whose obstetric history was unremarkable.

On examination, she was healthy looking with stable vital signs. There was a pfannanstiel scar that healed by primary intention. The symphysio-fundal height was 38cm compatible with her gestational age. There was a singleton fetus in longitudinal lie and breech presentation. The fetal heart tone was present and regular. There was no palpable uterine contraction.

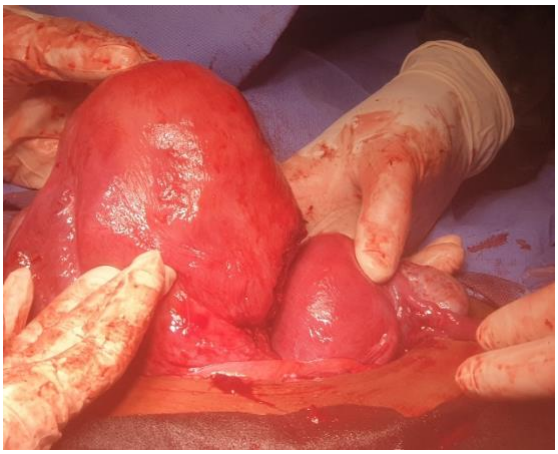

Figure 1: ??? Title

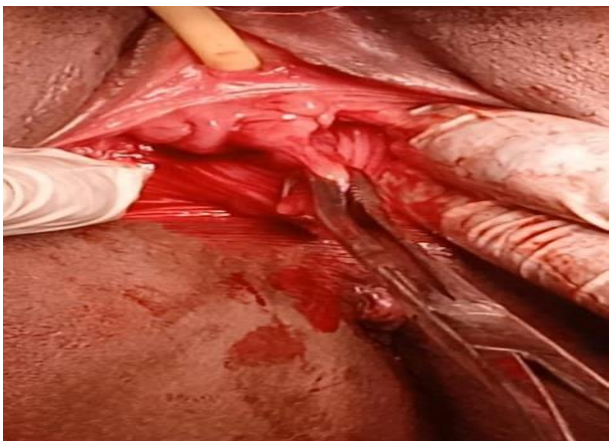

Figure 2: ??? Title

Her preoperative investigations were normal and she had two units of blood grouped and cross matched.

The intraoperative findings were that of a mild pelvic adhesion with a well-formed lower uterine segment. Liquor was clear and a live male fetus was delivered by breech extraction. The APGAR scores were 8 and 9 in the first and fifth minutes respectively, and the birth weight was 3.4kg. the placenta was posteriorly implanted. The uterus was repaired in two layers. It was discovered that the uterus had only one ovary and fallopian tube. Upon further exploration, a second fully developed bulky uterus was discovered below and to the right of the previously pregnant uterus (figure 1). Completely separate from it. On it was attached a well-developed ovary and fallopian tube. On further exploration, a longitudinal vaginal septum was discovered separating the 2 uteruses each with a grossly normal well-developed cervix (figure 2).

She did well and was discharged home. She had and IVU six weeks postpartum which revealed a right renal diverticulum.

Case B

She was an 18year old unbooked primigravida who presented in active phase of labour. The pregnancy was spontaneously conceived and had remain uncomplicated. She had no history of dysmenorrhea or dyspareunia. She had no prior medical or surgical history of note.

She had a spontaneous vaginal delivery of a healthy life male neonate with APGAR scores of 8 and 9 in the first and fifth minutes respectively and a birth weight of 3.1 kilogram.

She later developed primary postpartum haemoarrhage due to what was thought to be and endometrial poly. She had additional oxytocic, misoprostol and uterine massage. She also had 3 doses of 1gram of tranexamic acid. The bleeding stopped and she had two unit of blood transfused. She did well and was discharged home to be seen in 6 weeks for a planned polypectomy.

She presented as scheduled and was prepared for a polypectomy under subarachnoid block. Intra operative findings was that of a normal vulva and vagina. There was a polyp like mass protruding from the uterus via the cervical Os with it stalk deep within the uterine cavity. In the process of excising the poly, an ovary was discovered within it prompting the suspicion of an inadvertent uterine rupture. This necessitated an abdominal exploration where the uterus was found to be normal with no breech or abrasion on its wall. However, a dimple was found on the right side of the apparently grossly normal uterus. This was continuing with the stalk of the "polyp" (Figure 3). The polyp was pushed back into the abdomen and reoriented. It was then discovered that it was uterine didelphys that got inverted during her last child birth causing primary postpartum haemorrhage (Figure 4).

She had subtotal hysterectomy of the inverted extra uterus. She had an IVU done which was normal. She did well and was discharged home.

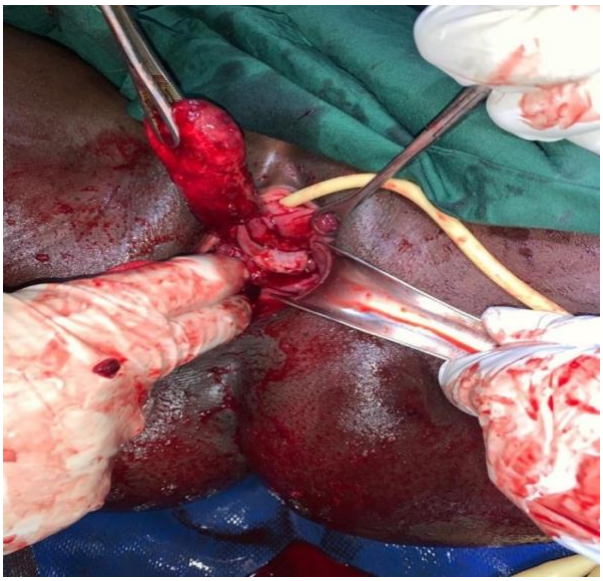

Figure 3: Title???

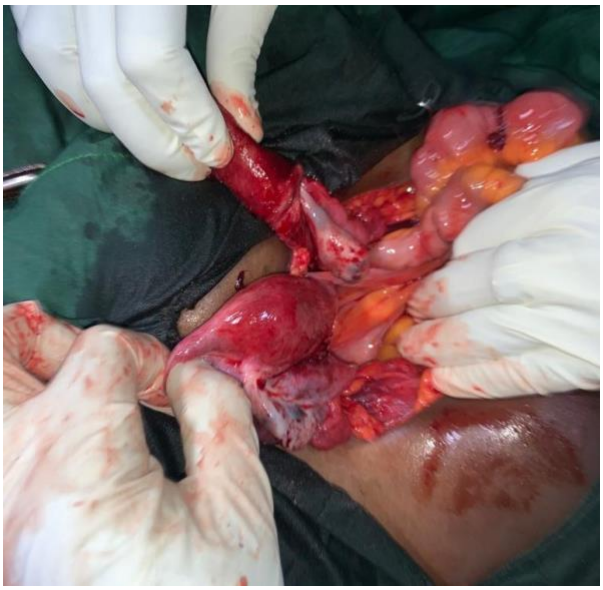

Figure 4: Title???

Discussion

Here we present a 2-case series of uterine didelphys with two different mode of presentation, diagnosis and management. Uterine didelphys is rare, seen in about 8-10% of all uterine anomalies and 0.3% in general population and higher among infertile patients.[11] Though modalities exist for its early diagnosis, It is often discovered incidentally as seen in these case series. This is owing to its asymptomatic nature. Case A was discovered incidentally during her 3rd repeat elective caesarean section, while Case B was discovered during what was thought to be a polypectomy. They both had no prior symptoms or signs that may prompt evaluation and discovery. Case A had persistent breech presentation in all of her pregnancies including the index pregnancy. Malpresentation is a common complication seen in women with uterine didelphys.[11] Breech presentation is reported in about 43% of uterine didelphys.[5] While case B had a singleton gestation in cephalic presentation. Other complications of uterine didelphys such as dysmenorrhea, dyspareunia, infertility, miscarriage, preterm labour and intrauterine growth restriction were not seen in both cases. The risk of caesarean section is increased in women with uterine didelphys.[7] This may be as a result of preterm labour, intrauterine restriction, malpresentation or obstructed labour. Case A had caesarean section in all her deliveries. While case B had spontaneous vaginal delivery. Vaginal septum can be seen in association with uterine didelphys in about 70% of cases and renal anomaly in 40% of mullerian anomalies including didelphys.[3, 5, 12] Complete longitudinal vaginal septum and renal diverticulum was seen in case A but none was discovered in case B. Post-partum haemorrhage may follow delivery in uterine didelphys. This was not seen in case A, However, it was the case in case B, which lead to the discovery of what was thought to be an endometrial polyp. It was later discovered to be an acute uterine inversion of the extra non pregnant uterus. Preterm delivery and intra uterine restriction are other documented complications. These were not found in both cases presented, as the pregnancies were both carried to term and babies were of average sizes. Fetal survival rate in uterine didelphys was found to be around 75% of the 49 cases of women with uterine didelphys evaluated by Heinonen.[7] All the fetuses in this series survived with no apparent complication.

Conclusion

Uterine didelphys is a rare congenital uterine anomaly with no specific symptoms. This made most of its diagnosis incidental. It may be associated with infertility, recurrent pregnancy loss, fetal malpresentation and adverse pregnancy outcome. As such, it should be part of the evaluation offered to a patient with such presentation. Patient diagnosed with uterine didelphys should be counselled on implication and have modality of management discussed with them. Psychological support should also be provided.

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