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Congenital Uterine Anomaly: An Incidental Diagnosis-Report of Two Interesting Cases

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Abstract

Woman with uterine malformations have higher rates of reproductive loss, pre-term delivery, infertility, intra uterine growth retardation, breech presentation and complications that increase obstetric intervention and perinatal mortality. The best way of diagnosing these anomalies is by using imaging techniques. We present a case where in full term pregnant woman was diagnosed to have congenital uterine didelphus following delivery of female baby with vertex presentation by LSCS (lower segment caesarean section) and an other case of uterine anomaly in a young woman with recurrent pelvic pain. Both the cases were diagnosed incidentally on gross examination of specimen following hysterectomy and which was not diagnosed initially by ultrasound scan.

Keywords: Mullerian anomalies; Recurrent pregnancy loss; Pregnancy-induced hypertension; Accessory and cavitated uterine mass

Introduction

The uterus is formed during embryogenesis by the fusion of the two paramesonephric ducts (also called mullerian ducts). This process usually fuses the two mullerian ducts into a single uterine body. Incomplete fusion of the mullerian ducts results in uterine malformation like uterus didelphus, uterus bicornis bicollis, uterus bicornis unicollis, uterus subseptate, uterus arcuate, and uterus unicornis. Uterus didelphus is less common than other uterine malformations and has been estimated to occur in 1/3000 women. It represents a uterine malformation where the uterus is present as paired organ due to embryonic non-fusion of the mullerian ducts. Each uterus has a single horn linked to the ipsilateral fallopian tube which faces its ovary [1].

Case Report

Case 1

20 years female Gravida 3 Abortion 2 (G3A2), with full term pregnancy presented with pain abdomen of 2 days duration. She appreciated fetal movements well. There was no history of leaking, bleeding per vagina, headache or blurring vision. She had bad obstetric history with two spontaneous abortions at 3 months of pregnancy for which dilatation and curettage was not done. The present pregnancy was uneventful except for the above symptoms. On Physical examination pallor was present. Blood pressure was 150/90 mm Hg, pulse-70 beats/min, Hb-8.4 gm% and her blood group was B positive. Per-abdomen examination revealed uterus of 34 weeks, active with, 1-2 contractions for 15-20 seconds for a period of 10 minutes and cephalic presentation. Fetal heart sound was 132 beats/min. Per-vaginal examination showed two vaginal openings separated by longitudinal vaginal septum, two cervices, one of the cervixes was 2 cm dilated with good effacement and vertex presentation at -1 station, membranes were intact, and pelvis was adequate. Biochemical investigations were within normal limits. Ultrasonography revealed single live intrauterine pregnancy of 34 weeks ± 5 days gestation in cephalic presentation. Final clinical diagnosis was G3A2 with term pregnancy, vertex presentation and longitudinal vaginal septum in active labour with fetal distress (thick meconium stained liquor). LSCS was done for fetal distress. On peroperative findings, live female baby weighing 2.4 kilogram was extracted by Patwardan's technique. Baby cried immediately after delivery and Apgar score was 9. Liquor was thick and meconium stained. Placenta and membranes was expelled in toto. Atonic postpartum haemorrhage was present. Hence bilateral uterine artery ligation, bilateral B lynch suture, bilateral internal iliac artery ligation was done followed by subtotal hysterectomy. Post-operative period was uneventful. Specimen was sent for histopathological examination. Specimen consisted of two separate uterine cavities (Figure 1). Larger uterus measured 15 cm×10 cm×4 cm and smaller uterus measured 7 cm×5 cm×4 cm. Cut section of both uterus showed endomyometrial thickness of 2 cm. Sections studied from both uterus showed hyperplastic myometrium with decidual lining (Figure 2). There was no evidence of chorinoic villi.

Case 2

32 year female, para 1 and living 1 presented with pain abdomen and backache of 4 months duration. Per-abdomen was soft, with mild

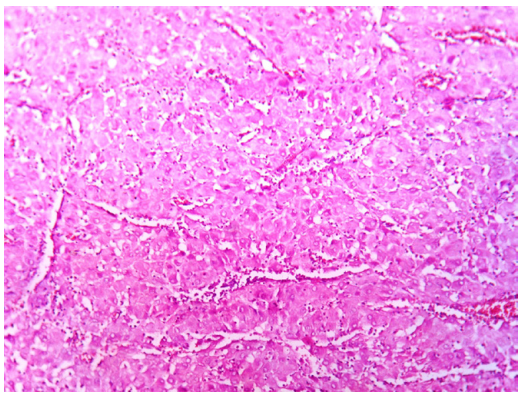

Figure 1: Gross photograph of Uterus didelphus (arrow) (case 1).

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Figure 2: Microphotograph showing endometrium with decuidalization. No chorinoic villi seen. (H&E, ×10) (case 1).

Figure 3: Gross photograph showing accessory and cavitated uterine mass on left side (arrow) (case 2).

tenderness in left iliac fossa. On per-speculum examination, cervix and vagina was healthy. Per-vaginal examination revealed bulky uterus, which was retroverted and deviated to right. Ultrasound scan was done and reported as pelvic inflammatory disease with 6.2 cm×3.8 cm tubo-ovarian mass in left adnexa. Clinical diagnosis of chronic pelvic inflammatory disease with tuboovarian mass was made. Patient underwent total abdominal hysterectomy with bilateral salpingo-oopherectomy. Grossly there were two uterine cavities as shown in (Figure 3). On microscopic examination, endometrium was in proliferative phase in both uterine cavities. Cervix, bilateral fallopian tubes and ovary were within normal limits and there was no tubo-ovarian mass. Diagnosis of non communicating accessory and cavitated uterine mass (ACUM) was made.

Discussion

The true incidence of congenital uterine anomalies in the general population and among woman with Recurrent Pregnancy Loss (RPL) is not known accurately. Although incidences of 0.16 to 10% have been reported, the overall data suggest an incidence of 1% in the general population and 3% in woman with RPL and poor reproductive outcomes. Overall, the prevalence of major congenital anomalies appears to be three-fold higher in woman with RPL compared with woman without a history of recurrent miscarriage [2]. Many nonobstructing uterine abnormalities are asymptomatic and may be discovered only in the evaluation of RPL, persistent menstrual irregularities or infertility. The best way of diagnosis is by using imaging techniques. Additional complicating matters include the lack of uniform imaging modalities for diagnosis [3,4].

Acien studied 176 patients with uterine malformations including bicornuate (n=49), didelphus (n=15), septate uterus (n=17) and 28 woman with other genital and/or urinary anomalies but with a normal uterus. It was reported that patients with uterine malformations have higher rates of reproductive loss, pre-term delivery, infertility, intra uterine growth retardation, breech presentation and complications that increase obstetric intervention and perinatal mortality [5]. In our first case patient presented with recurrent abortions and uterine anomaly was not diagnosed initially.

Ben-Rafael et al. evaluated the incidence of pregnancy induced hypertension (PIH) in woman with congenital uterine malformations by examining the pregnancy complications of 67 women with uterine anomalies compared with a control group of 130 women with normal-shaped uterus. He reported a significantly increased (p less than 0.04) rate of PIH in woman with uterine malformation as well as a 2-fold higher frequency of preeclampsia. In our first case patient also had PIH [6]. Ludmir et al. conducted studies for 8 years managing 42 women swith 101 pregnancies with previously diagnosed but uncorrected uterine malformations referred to the institution for high-risk obstetric care. The population studied consisted of 4 groups of pregnancies with uterine anomalies as unicornuate [5], bicornuate (61), septate (25), and didelphus (10). Sixty percent of the pregnancies in the unicornuate and didelphus group reached term, whereas it was 39% in the bicornuate and 48% in the septate group [7].

The recommended surgical technique is to unify the uterus which is the method of Strassman [8]. In one report, eight patients with uterus didelphus and recurrent abortion underwent Strassman metroplasty. Four of the five patients with follow-up information had living children postoperatively [9]. Because there are only anecdotal reports and no randomized studies, surgical metroplasty should be reserved, on a case-by-case basis, for selected patients who suffer from RPL or premature births [10].

The cavitated accessory uterine mass with functioning endometrium is a new type of mullerian anomaly in women with an otherwise normal uterus. This entity is problematic because of a broad differential diagnosis, including rudimentary and cavitated uterine horns; and is generally under diagnosed, being more frequent than previously thought [11]. In the literature searched ACUM's with otherwise normal uterus have been reported in young women with severe dysmenorrhoea and chronic/recurrent pelvic pain as seen in our case 2. ACUM is located in the anterior wall of the uterus at the level of insertion of the round ligament.

It presents with a certain similarity with the cavitated true adenomyomas observed in older woman in whom the endometrial lining of the cystic cavity is generally absent. For differential diagnosis with cavitated noncommunicating rudimentary uterine horns, hysterosalpingography showing a normal eutopic uterine cavity is decisive [12]. ACUM's could be caused by duplication and persistence of ductal mullerian tissue in a certain area at the attachment level of the round ligament, possibly related to a gubernaculums dysfunction. Early surgical treatment involving the laparoscopic or laparotomic removal of the mass could prevent the usual prolonged suffering of these young women [11]. In our opinion, this entity is a new mullerian anomaly.

Conclusion

Congenital uterovaginal anomalies can have adverse effects on pregnancy outcome. Early diagnosis and an aggressive evaluation of any patient presenting with mid-trimester abortion, premature labour, malpresentation, or retained placenta may prevent pregnancy

wastage and maternal morbidity. With timely and accurate diagnosis, appropriate management is likely to provide the best possible outcome for all such patients.

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