

Rudimentary Horn Pregnancy- A Diagnostic Dilemma

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ABSTRACT

Pregnancy implanted in the rudimentary horn of unicornuate uterus is very rare with an incidence of 1 in 75,000 to 1 in 150,000 pregnancies and 50% of them rupture, while 80% of rupture occurs in first and second trimesters, where atrocious outcomes are reported. Horn pregnancy rarely reaches third trimester and when it does the diagnosis becomes more challenging. Ectopic in unicornuate rudimentary horn of uterus is difficult to diagnose even with best facilities available. Consequences are grave with slightest error of radiological and clinical judgment.

Here we report a case of a 20-year-primigravida with 34 weeks, IUD baby who reported to labor ward with pain lower abdomen. She was induced and in the process she went in to shock following which laparotomy was performed and per operatively we found ruptured rudimentary horn, and unilateral horn resection with salpingectomy was performed. The purpose is to spread awareness of all such rare cases where maternal and fetal well is jeopardized, and accentuates the significance of high index of suspicion in all such cases.

Keywords: Ectopic; Rudimentary horn; Mullerian anomaly; Rupture

INTRODUCTION

Mullerian anomalies were first classified in 1979 by Buttram and Gibbons which was revised by the American Society of Reproductive Medicine in 1988. Developmental deviations can result in Unicornuate uterus with rudimentary horn [1-3]. ASRM classification places unicornuate uterus in to type-2 classification with unilateral hypoplasia or agenesis which is further classified into communicating, non-communicating, no cavity and no horn [4]. Its incidence is 2.4-13% of all the Mullerian anomalies [5]. 72-85% of the rudimentary horns are non-communicating with the cavity [6]. Various gynecological and obstetric complications like infertility, hematometra, urinary tract anomalies, abortions, preterm deliveries are associated with unicornuate uterus with rudimentary horn. Pregnancy in a rudimentary horn is a very rare condition with an incidence ranging from 1 in 75000 to 150000 pregnancies and results in uterine rupture 50-90% of the times mostly by the end of second trimester [7,8]. Diagnosis is difficult as majority of cases present in emergency with hemoperitoneum in early trimesters. Here we report a case of ruptured rudimentary horn pregnancy of 34weeks gestation which was misdiagnosed as intrauterine pregnancy with fetal demise with abruption.

CASE PRESENTATION

20 years primi with 34 weeeks of gestation reported to the emergency with chief complaints of labour pains since 6hours. She came with a USG report showing intrauterine fetal demise of 34 weeks gestation. On general examination, her blood pressure was 150/96 mm hg and pulse rate 110/min. Urine albumin was traces. On per abdomen examination, uterine size was 30 weeks and fetal heart sound was no localized. Uterus was tense and tender. On per vaginal examination, OS was

Received: 15-December-21	Manuscript No: IPGOCR-21-12004
Editor assigned: 17-December-21	PreQC No: IPGOCR-21-12004 (PQ)
Reviewed: 12- January -22	QC No: IPGOCR-21-12004
Revised: 19- January -22	Manuscript No: IPGOCR-21-12004 (R)
Published: 26- January -22	DOI: 10.21767/2471-8165.100001

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Citation: Khanam D, Hakim S, Mhatre S (2022) In Gynaecologic Oncology, Research on Cancer Care Delivery is Being Conducted. Gynecol Obstet Case Rep. Vol.8 No.1:1.

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closed with uneffaced cervix. No bleeding was seen. A diagnosis of abruption with fetal demise was made and patient was induced with Tab Misoprost 50 microgm S/L 4 hourly according to bishops' score with maximum of five doses. After completion of five doses, Inj Syntocinon was started 5 units @ 10 dpm and was titrated according to pains. After 6 hours of syntocinon, patients' blood pressure started falling with tachycardia. USG guided tapping was done and blood was aspirated from peritoneal cavity. Patient was immediately shifted to OT for Laparotomy in view of rupture uterus.

Intraoperatively, around 1.5 L of hemoperitoneum was found with left sided ruptured non- communicating rudimentary uterine horn of a unicornuate uterus as shown in **Figures 1 and 2** and intact sac with male fetus still lying inside the rudimentary horn. The weight of the fetus shown in **Figure 3** was 1.5



Figure 1: Ruptured site over left rudimentary horn.



Figure 2: Communication between unicornuate uterus and left rudimentary horn.



Figure 3: Male dead baby with skin peeling.



Figure 4: Resected left rudimentary horn with ruptured site.

kilograms. The left sided rudimentary horn was removed along with left sided salpingectomy leaving left sided ovary intact as shown in **Figure 4.** After achieving homeostasis, abdomen was closed in layers and drain was put in-situ. 3 units of PRBC and 2 units of FFP were transfused. Patient was discharged on postoperative day 10 after stitch removal in healthy condition.

DISCUSSION

Failure of the complete development of one of the Mullerian ducts and incomplete fusion with the contralateral side results in the development of a rudimentary horn with a unicornuate uterus. Mauriceau and Vassal in 1669 were the first to described pregnancy in a rudimentary horn [9]. Pregnancy in a non-communicating rudimentary horn is rarely observed and is thought to be due to transperitoneal migration of spermatozoan or fertilized ovum [10]. It is very rare to achieve a viable pregnancy in such cases and most of them result in rupture of the horn in first or second trimester of pregnancy [11]. Few cases reach term and fetal salvage is only 2% [12]. Prior to rupture diagnosis is difficult as sensitivity of USG to diagnose rudimentary horn pregnancy is around 26%-30% which further decreases with the advancement of pregnancy. In the present scenario the patient reported at 30 weeks but the scan could not diagnose it as a rudimentary horn, and was induced which resulted in rupture of the rudimentary horn. So the question is when to suspect a rudimentary horn pregnancy? The present experience supports the fact that any disproportionate pain in pregnancy should always be thoroughly evaluated. A high index of suspicion should be kept when a pregnant uterus does not respond to induction. MRI should be considered whenever inconclusive diagnosis is made on ultrasound [13-14]. A criteria for diagnosing pregnancy in rudimentary horn was outlined by Tsafir A, et al. [13]. They are: A) A pseudo pattern of asymmetrical bicornuate uterus, B) Absent visual connective tissue surrounding the gestation sac and the uterine cervix, C) Presence of myometrial tissue surrounding the gestation sac. However only few cases are diagnosed before rupture and mostly present as an emergency with hemoperitoneum. Cases of false and late diagnosis have also been reported. Rupture after use of misoprostol due to misdiagnosis has been reported by Samuels TA and Awonuga A [15]. A rudimentary horn pregnancy in 6th gravida was reported by Buntungu KA, et al. [16] with all previous normal vaginal deliveries.

Rudimentary horn pregnancy ultimately results in rupture and the gestational age at which it ruptures depends on the horn musculature and its distensibility, with 70-90% of rupture occurring before 20 weeks of gestation [17]. Although cases of pregnancy progressing to term and resulting in live birth after cesarean section has been documented [11]. Bleeding in rudimentary horn pregnancy rupture is more severe as uterine wall is thicker and more vascular [18]. A rudimentary horn pregnancy can also be complicated by placenta percreta with the incidence of 11.6% due to poorly developed musculature and the small size of the horn [19]. Hence the morbidity associated with it is very high.

Surgical removal is the primary strategy for managing rudimentary horn. Edelman AB, et al. [20] reported successful management of rudimentary horn pregnancy with methotrexate at an early gestational age. But with medical management further horn pregnancy cannot be prevented so mainstay is surgery [21]. However, in some cases with high resource setting, conservative management was attempted until the viability was advocated [22,23]. Prophylactic removal of non-communicating horn prior to pregnancy to prevent complications is also advised. Renal anomalies are associated in 36% of the cases; hence it is advised to further investigate these women for renal anomalies [21].

CONCLUSION

Despite the advances in imaging and diagnostic modalities, the antenatal diagnosis of rudimentary horn pregnancy is still difficult for clinicians. Delayed diagnosis or misdiagnosis can worsen the condition of the patient and golden period may be lost. All such cases with varied presentations of rudimentary horn pregnancy should be reported so that the clinicians are aware of atypical clinical scenario and maternal and fetal morbidities are avoided. A high index of suspicion for uterine anomalies should be made in early gestations and also in cases with failed induction. Early diagnosis of rudimentary horn pregnancy with early intervention reduces the maternal morbidity and mortality. With a rudimentary horn pregnancy, excision of the horn and ipsilateral salpingectomy is the recommended surgical treatment.een provided as evidence for this idea. E2 and P levels in the blood have also been found to be low in women who have had ectopic pregnancies. Do they represent the blastocyst's altered bioactivity of hCG or the creation of a faulty corpus luteum during ovulation? Could these low amounts of sex steroids play a function in modifying tubal transport of the fertilised egg, resulting in delayed embryo movement within the fallopian tube, because sex steroids have been found to play a role in oviductal motility? What about women who have no obvious risk factors but have an ectopic pregnancy? We previously showed the inaccuracy of a medical history by documenting evidence of a past pelvic infection through laparoscopy without the patient's knowledge. Antibodies to Chlamydia trachomatis have also been found in women who have had an ectopic pregnancy but have never had a STI, and evidence has been given suggesting antibodies to the C. trachomatis 70-kDa heat shock protein may play a role

in tubal mucosal injury pathogenesis.

Alternatively, investigations conducted by Blaudau in rabbits with extensive fimbria in their oviducts, similar to those found in humans, support aetiology of ectopic pregnancy that is unrelated to damaged fallopian tubes. They showed that the cumulus oophorus, which surrounds the ovulated egg as it travels from the ovary to the tubal ostium, is critical. Cumulus-free eggs showed delayed transit into the oviduct after enzymatic removal of the cumulus. The highly negatively charged gylcosaminoglycans in the cumulus interacted with the ciliated fimbria, assisting the egg's movement, they suggested. Ectopic pregnancy can develop in healthy fallopian tubes, and the lack of a previous STI does not rule out the possibility of a diagnosis. Knowledge of the risk factors linked to a higher likelihood of this diagnosis will not only lower the risk of rapid haemorrhage and death, but will also raise the likelihood of an earlier diagnosis, allowing for successful medical therapy without the risks and costs of surgery. Furthermore, if surgery is required, the fact that ectopic pregnancies can develop in normal fallopian tubes should provide justification for tubal preservation.

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