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Case Report

Successful diagnosis and management of prerupture rudimentary horn pregnancy in the second trimester: a case report *,**

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ABSTRACT

Rudimentary horn pregnancy has concerns due to the high incidence of an extreme risk of a life-threatening rupture. Thus, early diagnosis and management are essential to preserving the patient's life. We present a successful diagnosis and management of a prerupture rudimentary horn pregnancy in a 24-year-old woman presented with chronic pelvic pain and amenorrhea for the last 3 months. On physical examination, she had a mobile, nontender mass equals 16 weeks of gestation. Transvaginal ultrasound revealed an empty uterus with signs of a decidual reaction and a gestational sac adjacent to the uterus and surrounded by less than a 2 mm-in-thickness muscular wall with a positive fetal heart rate. The gestational age was 16 weeks based on biparietal diameter and femur length. Based on these findings rudimentary horn pregnancy was suspected. Laparotomy was performed, unicornuate uterus with unruptured, left rudimentary horn pregnancy was observed, and the pregnant horn with the ipsilateral tube was excised. To conclude, an empty uterus and extrauterine gestational sac surrounded by a thin muscular wall (<2 mm) on ultrasound should raise the suspicion of rudimentary horn pregnancy.

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Introduction

A unicornuate uterus is a rare congenital anomaly that arises when one of the müllerian ducts fails to develop; partial development resulted in a contralateral rudimentary horn [1]. Several women remain asymptomatic, which leads to an incidental diagnosis. Another subset of patients may experience chronic abdominal pain or dysmenorrhea [2].

The most significant concern is the probability of implantation in the rudiment horn and its sequential complication. The incidence of rudimentary horn pregnancy ranges from

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Fig. 1 – Shows an empty uterus with signs of decidual reaction.



Fig. 2 – Exposes a thin muscular (<2 mm) surrounding the gestational sac.

1 per 76,000 to 1 per 140,000 pregnancies [3]. And rupture of the pregnant horn is the leading cause of death and arises in approximately half of cases [4,5]. Because patients often have a history of prior normal pregnancies [4], and 80%-90% of rudimentary horn pregnancy ruptures early in the second trimester [6,7], both early diagnosis and delivery of pregnancy are crucial for decreasing maternal mortality rate [4,7].

Herein, we present a successful diagnosis and management of 16 weeks, prerupture rudimentary horn pregnancy.

Case presentation

A 24-year-old women gravida 3 para 2 presented to our department with a dull, continuous pain in the lower abdomen. She stated that the pain started a month ago and did not respond to analgesics. She also said that she was missing her menstrual period for the last 3 months in addition to episodes of vaginal spotting. The patient had a history of 2 full-term pregnancies that were delivered vaginally, and the final one was a year and a half ago. The remaining of her gynecological, medical, surgical, and familial history was unremarkable. She was afebrile and appeared slightly paled. Her vital signs were stable. Physical exam showed a mass on the left side of the umbilicus consistent with a 16-week gestation. Vaginal inspection showed a closed cervix. Bimanual examination revealed a mobile, nontender mass that extended to the umbilicus and seemed separate from other surrounding organs. Lab results revealed mild anemia and positive urinary β -human chorionic gonadotropin. Liver and renal function tests were within the normal range, as well as PT and PTT.

The transvaginal ultrasound exposed an empty uterus with signs of decidual reaction extending to 1.7 cm (Fig. 1:). We detected a gestational sac surrounded by a thin muscular wall (less than 2 mm) next to the uterus (Fig. 2:) with a positive fetal heart rate. We estimated the gestational age to be around 16 weeks based on the biparietal diameter and the

femur length, 12 mm and 21.5 mm, respectively. Renal abnormalities were excluded by ultrasound.

We set abdominal pregnancy and rudimentary horn pregnancy in our differential diagnoses. Although MRI can provide more details about the ectopic pregnancy, it was unavailable in our hospital, and the patient was unable to afford the high cost of MRI in a private center. Obtaining the MRI would not have changed the necessity for laparotomy in the management of the patient.

After discussing the life-threatening risks of continuing the pregnancy, our patient provided her written informed consent to terminate the pregnancy. She underwent laparotomy under general anesthesia, which revealed a unicornuate uterus held by a singular round ligament and 2 uterosacral ligaments, and it had a rudimentary horn with one fallopian tube with a round ligament and no cervix. The gestational sac was within the rudimentary horn (Fig. 3:), which had a vascular wall and no connection to the uterus. We resected the rudimentary horn completely with the ipsilateral fallopian tube (Fig. 4:), and closed the abdomen's layers. She did not require a blood transfusion. We used the ultrasound to investigate the kidneys looking for abnormalities; both kidneys were in their normal anatomical position. She was discharged 48-hour postoperation.

After 2 weeks, the patient came for a follow-up visit. She was in good condition and had no further complications.

Discussion

A unicornuate uterus is a congenital anomaly resulting from the failure of one of the müllerian ducts to develop [6]. It represents 4%-5% of uterine abnormalities [5,8] and further classified into 4 categories: (1) communicating, cavitary, rudimentary horn, (2) noncommunicating cavitary, rudimentary horn (3) noncavitary rudimentary horn (4) Unicornuate uterus without a rudimentary horn, which is the most common form



Fig. 3 – The figure reveals a left pregnant rudiment horn (black arrow) and the uterus (blue arrow).

[2,4]. Over half of the rudimentary horns comprise a cavity with functional endometrium, and 72%-80 % of them are noncommunicating [4,5,8]. Most functional noncommunicating horns present during or after the third decade of life [4]. Although the right-sided rudimentary horn usually rampant in 54% of cases [5], our patient had a left-sided rudiment horn which relatively unusual.

The isolated form of the unicornuate uterus is usually asymptomatic and identified incidentally. On the other hand, the clinical picture varies when it is associated with a rudimentary horn. Patients often present with chronic pelvic pain and dysmenorrhea after menarche or later in life [2]. Most importantly, pregnancy can occur in the rudimentary horn leading to severe complications such as abortion, prematurity, intrauterine growth retardation, and fetal death [4,5]. The implantation is more frequent in the noncommunicating cavitary horn [3]; this is illuminated by transperitoneal migration of the gametes [5].

Although cases of a full-term live fetus [9] and prerupture diagnosis and management of rudimentary horn pregnancy [3,10–13] have been reported, and the maternal mortality rate declined from 5.1% to lower than 0.5% [5], the outcomes remain poor and rupture of the pregnant horn – due to the defective musculature – is the leading cause of death and arises in about half of cases [4,5]. Since patients usually have a history of preceding conventional pregnancies [4], and the fact that



Fig. 4 – Shows the excised rudiment horn with fallopian tube and the fetus with the placenta.

80%-90% of rudimentary horn pregnancies rupture between the 10th and 15th weeks gestation [6,7], early diagnosis and delivery of pregnancy emerges as a challenge to decrease the maternal mortality rate and increase the live birth rate [4,7].

Tsafrir proposes 3 fundamental criteria on ultrasound for the ultimate diagnosis of pregnancy in a rudimentary horn, which are: pseudo-pattern of an asymmetrical bicornuate uterus; absent visual continuity between the cervical canal and the lumen of the pregnant horn; the presence of myometrial tissue surrounding the gestational sac [3]. However, the sensitivity of ultrasound is relatively low, extending from 29% to 33% [14]. Furthermore, MRI may provide thorough information about Müllerian anomalies and many cases of ectopic pregnancies [11,12], it was unavailable in our hospital and the patient was incapable of affording the high cost of MRI in a private center. It is worth mentioning that MRI results do not change the management protocol [12].

It is mandatory to differentiate rudimentary horn pregnancy from any other type of pregnancies [8]. Intrauterine pregnancy is characterized by a broad interaction along the uterine cavity and the gestational sac; this feature is not present in ectopic pregnancies [8]. Diagnosis of tubal pregnancy is made by ensuring that either a heterogeneous or noncystic adnexal mass is isolated from the ovary [15]. Interstitial pregnancy is diagnosed during the first trimester based on the following ultrasonographic findings: an external gestational sac separated from the lateral border of the uterus and surrounded by a thin, asymmetric myometrial layer, and an empty uterine cavity [16]. Diagnosis of intra-abdominal pregnancies is crucial and challenging as they are secondary to the rupture of a tubal pregnancy [8,15]. They favor the broad ligament [8,15], resulting in a deep, immobile pelvic mass on palpitation [8]. Ultrasound reveals an empty uterine cavity and a gestational sac separate from the uterus, fallopian tubes, and ovaries [15]. Moreover, the well-known association of abdominal pregnancy with intrauterine growth restriction and oligohydramnios; both of these findings cannot be seen in rudimentary horn pregnancy [8]. However, the definitive diagnosis may not be achievable until performing laparotomy, in our case for instance.

Removal of the rudimentary horn and the ipsilateral tube through either laparotomy or laparoscopy is the gold standard for management, whichever the trimester is [7]. It is recommended because the functional endometrial horn is associated with an increased risk for dysmenorrhea, infertility, or ectopic pregnancy. Moreover, the risk of rupture in the second trimester is very high [6,14].

The unicornuate uterus is usually associated with renal anomalies, contralateral renal agenesis being the most common anomaly [1,2]. Horseshoe kidney, unilateral medullary sponge kidney, and double renal pelvis are infrequent. As a result, whenever a unicornuate uterus is identified, evaluation of the renal system is warranted either by ultrasound, computed tomography, or magnetic resonance imaging [1]. In our case, we used the ultrasound to explore any renal abnormalities, which revealed bilateral kidneys in their normal anatomical position.

Conclusions

Implantation in the rudiment horn is a rare condition but has severe implications on fetal and maternal life. Thus, ultrasonographic findings such as an empty uterus and extrauterine gestational sac surrounded by a thin muscular wall (<2 mm) should raise the suspicion of rudimentary horn pregnancy.

Patient consent

Written, informed consent for publication of this case was obtained from the patient.

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