

Diagnosis and Management of a Ruptured Corpus Luteum Cyst: A Case Report from a Resource-Limited Setting

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ABSTRACT

Corpus luteum cysts are common in women of reproductive age and especially in the setting of pregnancy. More often than not it poses a challenge in diagnosis as it is confused for ectopic pregnancies especially when it ruptures and sometimes when it coexists with ectopic, heterotopic and intrauterine pregnancies. In low-resource settings, the unavailability of laparoscopic services, demands that exploratory laparotomy be done for diagnoses and further management. In this case report, a female patient presenting with acute abdomen symptoms underwent exploratory laparotomy for a ruptured corpus luteum cyst in the setting of intrauterine pregnancy which was aborted before term. This had been diagnosed with sonography as a case of heterotopic pregnancy. The addition of laparoscopy to sonography and β HCG should be the cornerstone of diagnosis and would stipulate further patient management.

Keywords: Corpus luteum; Ovarian cyst; Heterotopic; intrauterine; laparoscopy; ruptured; hemoperitoneum; β HCG.

INTRODUCTION

Previous studies reveal that 20% of women will develop pelvic mass during their lifetime [1]. Globally, about 7% of women have been reported to have ovarian cysts during their lifetime [2] However, the actual prevalence of ovarian cysts is unknown. They are common in women of reproductive age where they are majorly physiological cysts such as corpus luteum cysts or follicular cysts. They have also been reported among post-menopausal women however the risk of them being malignant cysts is reported to peak with increasing age [1]. Ovarian hyperstimulation, tamoxifen uses and pregnancy among others, have been listed as some of the risk factors for ovarian cyst formation [1]. Sometimes complications such as ovarian torsion, haemorrhage and rupture could occur [1].

After ovulation, the remnants of the follicles develop into the corpus luteum which produces progesterone to support the pregnancy in case the ovum had been fertilized. The corpus luteum will normally sustain the pregnancy for about 14 weeks, after which dissolution is expected to occur. If it does not occur, corpus luteum cysts will form and grow to about 3 cm. Corpus luteum cysts are a common occurrence in pregnancy, and are mainly asymptomatic and tend to end by the end of the first trimester [1].

Ruptured corpus luteum cysts in women of reproductive age can significantly pose tremendous diagnostic and management challenges to medical practitioners. More often than not, a ruptured corpus luteum cyst especially in the setting of early intrauterine pregnancy has been confused for a ruptured ectopic pregnancy or even heterotopic pregnancies in other instances [3]. Management can sometimes be an uphill task because these patients present with signs of acute abdomen, and in low-resource settings where a

laparoscopic examination is not possible, exploratory laparotomy come in handy to rule out our suspicion.

We report a case of a young female patient presenting with a ruptured corpus luteum cyst in early pregnancy that was confused for a ruptured ectopic pregnancy. The removal of the adnexal mass, which turned out to be histologically the corpus luteum resulted in the pregnancy loss.

CASE REPORT

A female patient aged 28 with a period of amenorrhea of about 43 days presented with a history of severe cramping abdominal pain that had lasted for about a week but worsened in the last 3 days before her hospital visit. She reported that the pain would come at an interval of about 5 minutes after which she would be fine once it subsided. Every incidence of pain was accompanied by the urge to empty her bladder and bowel and she had to remain still; in the position at which she was at the onset of the pain. She also reported painful micturition and a vague history of dyspareunia with vaginal soreness. She developed sudden onset post-prandial vomiting within our facility on that day. She had no history of family planning use. Her menarche was at 13 years. She had an irregular menstrual cycle and her menstrual flow would last four days. She reported dysmenorrhea majorly on her first day of every menstruation with was relieved by analgesia.

Clinical examination revealed the patient had no jaundice, no pallor, no lymphadenopathy, no dehydration and no cyanosis. She had a respiratory rate of 16 bpm, and she was tachycardic (105 bpm) with normal blood pressure (110/76) and normal oxygen saturation (96-98%). Abdominal examination revealed marked suprapubic tenderness, with no obvious distention.

A full hemogram showed normal haemoglobin levels (12.7g/dl), a normal white cell count ($6.7 \times 10^9/L$) and a normal platelet count ($360 \times 10^3/ uL$). The random blood sugar was 4.9mmol/L. Urine and stool analysis were normal. She had a positive β hCG test.

Transvaginal and transabdominal obstetric ultrasound revealed a single viable intrauterine pregnancy at 5 weeks 6 days and a right adnexal mass with abdominal and pelvic fluid that was concerning for a ruptured ectopic pregnancy (heterotopic ectopic pregnancy), Sub-serosal anterior myometrial fibroid in the lower uterine segment measuring 2.1×2.3×1.9cm and a large left lateral mass likely pedunculated myometrial fibroid measuring 4.4×4.7×5.9 cm.

The patient was scheduled for an emergency exploratory laparotomy. Intraoperative findings included a pedunculated fibroid measuring 8×7×2.5 cm for which a myomectomy was done. A ruptured right ovarian cyst measuring 3×2.6×1.7 cm, and a right cystectomy was done. There was a moderate amount of serosanguinous peritoneal fluid that was also suctioned. Bilateral ovaries were preserved.

Sections from the fibroid showed interlacing fascicles of smooth muscle fibres with no atypia, mitoses or necrosis. Foci of infarction and hyalinization were noted. Sections from the ovary showed minimal residual normal ovary with a cyst lined by luteinized cells whose cavity contains blood. There was no evidence of malignancy. Histopathological results concluded for the samples revealed leiomyoma uteri and luteum cyst respectively.

The patient was placed on antibiotics and analgesics postoperatively and recovered well until day 3 post op when she reported vaginal bleeding with dark clots, associated with severe lower abdominal pain. She was placed on 20mg of dihydrogesterone to minimize the bleeding for a transvaginal ultrasound to be done. The transvaginal ultrasound showed a non-viable pregnancy and the decision to terminate it with three cycles of 400mg misoprostol every 6 hours was made. Thereafter a repeat ultrasound showed complete evacuation. The patient was stable and was discharged home on oral antibiotics and analgesics.

DISCUSSION

Diagnosis of ruptured corpus luteum cyst in the setting of pregnancy can sometimes be challenging and on a few occasions has been confused for ectopic pregnancy. Bauman *et al.* presented a case of a 33-year-old woman with amenorrhea, positive β HCG, acute abdomen and a transvaginal ultrasound that showed hyperechogenic endometrium, 12 mm wide, with no gestational sac. A large amount of fluid was seen in the pouch of Douglas. A corpus luteum was seen in the left ovary and an undefined structure was suspected of ectopic pregnancy. Emergency laparoscopy revealed a ruptured corpus luteum, coagula and blood but no ectopic pregnancy. On day one postoperatively, transvaginal ultrasound was able to visualize an intrauterine gestational sac of 4 mm in diameter, and the pregnancy was carried to term delivery [3]. Vidakovic *et al.* presented a case like the above in a 35-year-old woman. A laparoscopy was performed and a ruptured luteum cyst was excised. Transvaginal ultrasound was able to visualize a 6mm gestational sac on postoperative day four. Progesterone therapy was started immediately after the surgery and the pregnancy was delivered at term uneventfully [3] [4]. A similar case was presented by Takeda *et al.* in a 15year old, presented with an acute abdomen and the gestational sac was visualized by the ultrasound together with hemoperitoneum. The preoperative differential diagnosis was narrowed to either intrauterine pregnancy with a ruptured corpus luteum cyst or heterotopic pregnancy. Emergency laparoscopic surgery showed a ruptured corpus luteum cyst whose bleeding site was sutured and hemostasis was achieved. The pregnancy was however terminated as per the request of the patient and the family [3] [5].

In our case report, the patient presented with amenorrhea, a positive β HCG, a transvaginal ultrasound that showed an intrauterine pregnancy and abdominopelvic fluid that was suspicious for a ruptured ectopic pregnancy (heterotopic pregnancy). Exploratory laparotomy revealed no active bleeding and no ectopic pregnancy was found. Since it was a wanted pregnancy, no curettage was done. Postoperative recovery was eventful with a miscarriage on the third day postoperative at 6 weeks gestation.

Acute abdomen in women of reproductive age in the setting of positive β HCG is always apprehensive for ruptured ectopic pregnancy, especially in the presence of hemoperitoneum, an adnexal mass, with an intrauterine pregnancy that cannot be visualized by sonography. In the setting of a negative pregnancy test, a ruptured corpus luteum cyst or other diagnoses such as appendicitis, ovarian torsion or endometriosis can be considered [6]. This generalization however may not be true in all instances as even with positive B HCG as in the setting of ectopic, heterotopic and intrauterine pregnancies, ruptured corpus luteum cysts have been mentioned [4]; with the cases presented in this case report serving as other examples.

In the case we presented, the intrauterine gestational sac was visualized and was a confirmation of a uterine pregnancy. This however was not possible with the other cases where the intrauterine pregnancy was at the earliest stages and thus only a thickened endometrium could be seen, which is not a reliable indicator of intrauterine pregnancy as this can also be seen with ectopic pregnancies and even the late luteal phase of the menstrual cycle. Thus, in pregnancies where the gestational sac is not visualized in the uterus, it is easier to conclude that there could be a preexisting ectopic pregnancy and a need to rule out our suspicion would be mandatory [4]. In the setting of an adnexal mass, hemoperitoneum and a visualized intrauterine pregnancy, as in our case, a conclusion of heterotopic pregnancy could easily be made sonographically.

In low-resource settings, like in our case presentation, exploratory laparotomy needed to be done to confirm our diagnosis and to manage the patient as laparoscopic services were out of reach. In stable patients with negative pregnancy test, management has been nonoperative. In acute presentations of ruptured corpus luteum cysts in pregnancy and with hemoperitoneum, laparoscopic explorations have been the gold standard [4]. According to Kim *et al.*, most cases of ruptured ovarian cysts with hemoperitoneum can be managed conservatively. Low diastolic blood pressure and a large amount of hemoperitoneum suggest the need for

surgical intervention [7]. From our discussion, compared to our case report, the pregnancies were carried up to term uneventfully. The miscarriage in our case could mainly be a result of delayed administration of progesterone hormonal therapy. Two case reports mentioned progesterone therapy immediately postoperatively in two of the pregnancies that were carried to term [4] [8].

Studies have shown non-operative management of adnexal masses to be preferred in many instances as the majority tend to regress during pregnancy and there is a high likelihood that it would be a corpus luteum cyst [9]. Removal of the corpus luteum cyst may reduce the level of progesterone which normally sustains the pregnancy in the first trimester before the formation of the placenta. Thus, progesterone should be administered to mitigate the risks of spontaneous abortions to patients who undergo surgeries within the first trimester according to the Society of American Gastrointestinal and Endoscopic Surgeons [10]. As corpus luteum cysts tend to regress by 12 to 16 weeks, watchful waiting can be done until after 16 weeks when the implantation is more secure as the corpus luteum would not be required to maintain the pregnancy after the first trimester [11].

Observation however should put into consideration the occurrence of acute events such as torsion or rupture which may require emergency surgeries [9]. If operative management is to be considered, elective surgeries would be preferred as they would bear minimal risks to the pregnancy, compared to emergency surgeries which have been associated with increased rates of miscarriage, premature rupture of membranes and preterm deliveries [9]. Our case was an acute presentation. Among other things, it had an ovarian mass in the setting of moderate peritoneal fluid which was worrying for ruptured ectopic pregnancy and thus emergency surgery, in this case, was inevitable. Were it possible, however, it would be advisable to delay the surgery to the second trimester or early post-partum. Laparoscopic surgeries are reported to have better surgical outcomes, nevertheless, no difference was reported in pregnancy outcomes between laparotomy and laparoscopic surgeries [9].

Surgical management of leiomyoma in pregnancy is discouraged except in rare cases and medical management is preferred in most instances [12]. Symptomatic pedunculated fibroids with a small stalk in pregnancy can be resected surgically [12]. In our case report, the fibroid was pedunculated and the patient consented to the myomectomy. The sub-serosal fibroid in the lower uterine segment was however not resected. Removal of the pedunculated fibroid was felt would not be associated with adverse events both for the mother and the baby. The pregnancy loss is mainly attributable to the removal of the adnexal mass at six weeks of pregnancy, as this was the corpus luteum. Leach et al, presented a case of successful pregnancy outcome post-first trimester myomectomy [13]. However, studies encouraging first-trimester myomectomies are still limited and thus management in such instances should be tailored to the individual patient [13].

CONCLUSION

To conclude, we should be conscious of ruptured corpus luteum cyst occurrence, with or without hemoperitoneum, coexisting with intrauterine or even ectopic pregnancy for females of reproductive age and laparoscopy should be employed in its diagnosis as a complementation to sonography and β HCG measurements and thereafter, therapeutic interventions can be done laparoscopically if available. Surgical interventions for adnexal masses should be discouraged within the first trimester unless they are emergent cases.

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