

Hemoperitoneum from corpus luteum cyst rupture in pregnancy of unknown location

Ali Babacan^{a,*}, Ismet Gun^a, Serkan Bodur^b, Yaşam Kemal Akpak^c, Murat Muhcu^a, Vedat Atay^a

^aDepartment of Obstetrics and Gynecology, GATA Haydarpaşa Training Hospital, Istanbul, Turkey

^bDepartment of Obstetrics and Gynecology, Maresal Çakmak Military Hospital, Erzurum, Turkey

^cDepartment of Obstetrics and Gynecology, Ankara Mevki Military Hospital, Ankara, Turkey

Abstract. The presence of corpus luteum cyst rupture resulting in hemoperitoneum together with a diagnosis of pregnancy at unknown location is rare and challenging for the gynecologist. A 29-year-old primipara woman presented with abdominal pain. A urine pregnancy test was positive. Ultrasonography revealed hemoperitoneum, but there was no sign of any intrauterine pregnancy. On abdominal exploration, there was a massive hemorrhage with a ruptured corpus luteum on the right ovary. The diagnosis and management difficulty of corpus luteum cyst rupture with a concomitant positive pregnancy test but without exact confirmation of pregnancy location is discussed.

Key words: Corpus luteum cyst rupture, hemoperitoneum, pregnancy of unknown location

1. Introduction

Corpus luteum cyst rupture resulting in hemoperitoneum is a rare clinical entity with an increased likelihood of rupture during pregnancy likely due to the increase in corpus luteum cysts in pregnancy (1-2). The clinical symptomatology and sonographic features of corpus luteum cyst rupture can closely mimic ectopic pregnancy (2). The diagnosis and management of hemoperitoneum due to corpus luteum cyst rupture could be extremely difficult for the gynecologist, especially when present with a pregnancy in an unknown location.

2. Case report

A 29-year-old primipara presented to the emergency room with a complaint of severe right lower abdominal pain for 4 hours. The laboratory findings of the patient were as follows: hematocrit 9.1%, prothrombin time 12s, activated partial thromboplastin time 25s, and urine

pregnancy test positive. Pelvic ultrasonography revealed massive hemoperitoneum (Figure 1a-b). The location of pregnancy could not be confirmed by either trans-vaginal or trans-abdominal ultrasonography. As the combination of acute abdominal pain, positive pregnancy test and hemoperitoneum is strongly suggestive of ruptured ectopic pregnancy, emergency exploratory minilaparotomy (≤ 3 cm) was performed. On exploration, there was a massive hemorrhage due to corpus luteum cyst rupture on the surface of the right ovary (Figure 2).

During the surgery, approximately 3L of hemorrhagic fluid was evacuated from the abdominal cavity. Hemostatic minimal electric coagulation was applied to the bleeding surface. Duration of surgery was 25 minutes. After surgery, luteal phase support with progesterone was performed. Thrombophilia panel and bleeding diathesis tests were ordered postoperatively due to abundant postoperative bleeding. Only remarkable finding was heterozygous positivity for MTHFR C677T. Ten days after laparotomy, a viable intra-uterine pregnancy was observed on ultrasonography and a 3150g healthy female baby was delivered at 39 weeks of gestation. The patient breastfed for 3 months and admitted to our outpatient department after 9 months with complaints of amenorrhea and flushing. Physical and laboratory findings suggested premature menopause: FSH, 92

*Correspondence: Dr. Ali BABACAN
GATA Haydarpaşa Training Hospital Uskudar 34668
Istanbul/Turkey
Tel: +90 532 3458254
Fax: +90 216 3487880
E-mail: ababacan_@hotmail.com
Received: 20.03.2014
Accepted: 04.09.2014

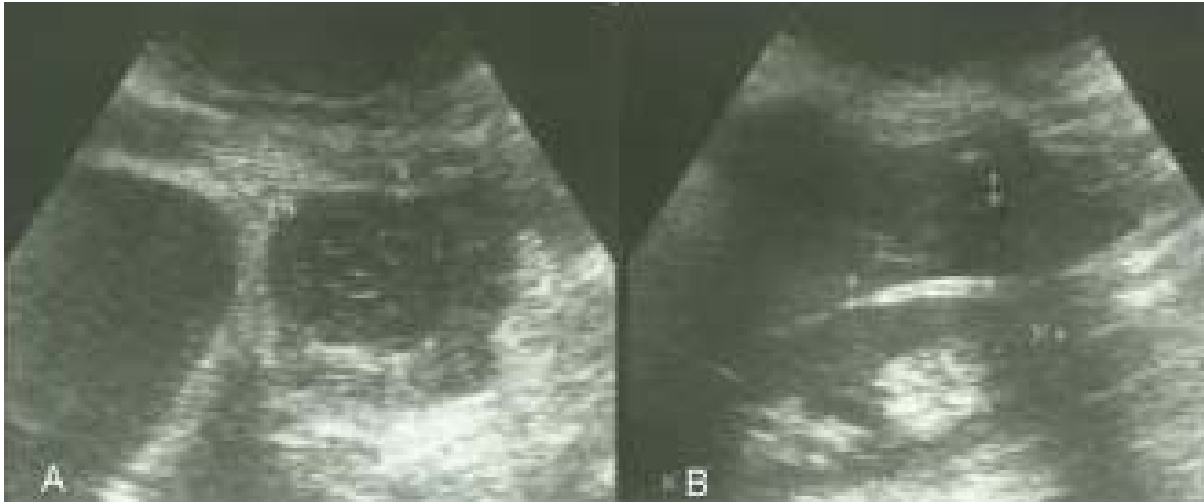


Fig. 1a). Ruptured corpus luteum cyst, b) Intraabdominal massive hemoperitoneum due to corpus luteum cyst rupture.



Fig. 2. Corpus luteum cyst rupture on the surface of the right ovary.

mIU/ml; E2 14 pg/mL. Corresponding hormone levels of 86 mIU/mL and 12 pg/mL after 6 months confirmed the diagnosis of premature menopause and hormone replacement therapy was initiated. Genetic workup revealed a normal karyotype of 46 XX.

3. Discussion

Hemoperitoneum is one of the causes of acute abdomen. Rupture of corpus luteum cyst is one of the major gynecologic causes of hemoperitoneum. Appropriate evaluation of gynecological hemoperitoneum always has a priority in practice because of the potential need for emergent surgical intervention.

Corpus luteum cysts are thin-walled, functional vascular structures, and most of them are predisposed to rupture (3). The etiology for cyst rupture is not known, although it has been suggested that the increased vascularity of the

ovary in the luteal phase and pregnancy may predispose to rupture of a corpus luteal cyst. The right ovary seems predisposed to rupture more than the left one, as seen in our case (4). One of the possible explanations for this predisposition is the protection of left ovary from trauma by the cushioning of the recto-sigmoid colon (1). By a thorough history after operation, we found that the abdominal pain had started immediately after sexual intercourse, highlighting the role of trauma in right-sided corpus luteum cyst rupture. Typically at the time of rupture, there may be sharp and sudden onset of pain, which has no typical characteristics. Blood loss can vary from very little bleeding to hypovolemic shock (1). At the time of operation, we found that the pelvic cavity was full of approximately 3L of hemorrhagic fluid. According to Hallatt et al (1) the hemorrhage from a ruptured corpus luteum cyst is likely to be less than in an ectopic pregnancy and likely to be non-recurrent once it stops.

Multiple sonographic patterns have been defined for hemorrhagic ovarian cyst and it has also been called the “great imitator” (5). Although the sonographic patterns of hemorrhage within an ovarian cyst have been well known, the findings related with cyst rupture and hemorrhage have not been well described in the literature (6). Consequently, patients with a ruptured ovarian cyst are frequently misdiagnosed with unrelated disorders (2). In one study, the hemoperitoneum was found to be the dominant imaging feature rather than the cyst itself (6). Hemoperitoneum from a ruptured hemorrhagic ovarian cyst exhibits imaging features similar to those of hemoperitoneum from other sources such as

ruptured ectopic pregnancy (7). In the current case, the hemoperitoneum was inconclusive for us in terms of the differential diagnosis between rupture of ectopic pregnancy and rupture of corpus luteum cyst. The diagnosis is mainly based on high clinical suspicion, laboratory data and ultrasound findings. Historically, the major differential diagnosis is ectopic pregnancy.

The diagnosis of corpus luteum cyst rupture is particularly difficult when it is presented with a positive pregnancy test because clinical and imaging findings can closely mimic ectopic pregnancy. In the literature, the main distinction between corpus luteum cyst rupture and ectopic pregnancy is based on a positive pregnancy test. Although, this kind of a generalization is true and practical in most of the cases, there are several case reports in the literature indicating that such assertions have only a limiting usage when those cases present together with a positive pregnancy test as in the current case. Furthermore, coincidental presence of corpus luteum cyst rupture with ectopic pregnancy has been reported (2, 5-6). Therefore, the possibility and occurrence of corpus luteum cyst rupture should be kept in mind even when the presence of intrauterine or extrauterine pregnancy is confirmed.

The corpus luteum cyst is a functional and hormonally active cyst that is required to maintain the pregnancy. In early pregnancy, it is formed immediately after fertilization of the ovum and it is responsible for progesterone production before placental production begins. It normally begins to regress around 8 weeks of gestation. Inadvertent surgical resection of the corpus luteum before 7 to 8 weeks may result in pregnancy loss (8). Prophylactic administration of progestational agents until placental maturation may be helpful in the event of inadvertent excision of the corpus luteum. In this case, the surgical intervention focused only on stopping bleeding by electro-coagulation instead of total excision, which played a crucial role in pregnancy continuation. Another factor in achieving a live fetus was the minimally invasive approach with mini-laparotomy. In our opinion, pregnancy and the patient gained benefit from minimal physiological and surgical stress caused by mini-laparotomy and short operative time.

CLH (corpus luteum hemorrhage) is a rare clinical entity, which can be attributed to hemorrhagic diseases. On the other hand, our case developed premature menopause shortly after the incident without any other cause. Our literature review did not reveal any publication addressing an association between CLH and premature menopause. In addition, the effect on

ovarian reserve of bipolar electrocoagulation is transient and causes minimal damage to ovary (9). In our patient's genetic analysis also revealed a normal karyotype. The patient did not also have a remarkable family history. She did not report smoking or substance abuse, previous surgery or chronic disease. Development of premature menopause after the operation, despite normal menstrual cycles and normal hormone levels preoperatively, suggests that CLH may be a possible cause of premature menopause. Further larger studies are warranted to clarify the issue.

Management of CLH is conservative or surgical (3,10-11). In patients with recurrent CLH, oral contraceptives are used to suppress ovulation (10). Ultrasonic evidence of large amounts of peritoneal fluid and severe pain is indication for operative intervention (3,10-11). In CLH, oophorectomy is rarely necessary (11). Minilaparotomic treatment of CLH of pregnancy in healthy women has not previously been reported. This form of minimally invasive surgery provides potential benefits for improved cosmetic appearance, shorter hospital stay, reduced postoperative pain, and earlier postoperative return to daily activities. In conclusion, ruptured corpus luteum cyst of pregnancy with massive hemoperitoneum should be considered in the differential diagnosis in the presence of a positive pregnancy test.

References

1. Hallatt JG, Steele CH, Jr, Snyder M. Ruptured corpus luteum with hemoperitoneum: a study of 173 surgical cases. *Am J Obstet Gynecol* 1984; 149: 5-9.
2. Raziell A, Ron-El R, Pansky M, et al. Current management of ruptured corpus luteum. *Eur J Obstet Gynecol Reprod Biol* 1993; 50: 77-81.
3. Hoffman R, Brenner B. Corpus luteum hemorrhage in women with bleeding disorders. *Womens Health (Lond Engl)* 2009; 5: 91-95.
4. Tang LC, Cho HK, Chan SY, Wong VC. Dextropleurancence of corpus luteum rupture. A clinical study. *J Reprod Med* 1985; 30: 764-768.
5. Jeffrey RB, Laing FC. Echogenic clot: a useful sign of pelvic hemoperitoneum. *Radiology* 1982; 145: 139-141.
6. Yoffe N, Bronshtein M, Brandes J, Blumenfeld Z. Hemorrhagic ovarian cyst detection by transvaginal sonography: the great imitator. *Gynecol Endocrinol* 1991; 5: 123-129.
7. Hertzberg BS, Kliewer MA, Paulson EK. Ovarian cyst rupture causing hemoperitoneum: imaging features and the potential for misdiagnosis. *Abdom Imaging* 1999; 24: 304-308.
8. Csapo AI, Pulkkinen MO, Ruttner B, Sauvage JP, Wiest WG. The significance of the human corpus luteum in pregnancy maintenance. I. Preliminary studies. *Am J Obstet Gynecol* 1972; 112: 1061-1067.

9. Özgönen H, Erdemoglu E, Günyeli I, Güney M, Mungan T. Comparison of the effects of laparoscopic bipolar electrocoagulation and intracorporeal suture application to ovarian reserve in benign ovarian cysts. *Arch Gynecol Obstet* 2013; 287: 729-732.
10. Payne JH, Maclean RM, Hampton KK, Baxter AJ, Makris M. Haemoperitoneum associated with ovulation in women with bleeding disorders: the case for conservative management and the role of the contraceptive pill. *Haemophilia* 2007; 13: 93-97.
11. Jamal A, Mesdaghinia S. Ruptured corpus luteum cysts and anticoagulant therapy. *Int J Gynaecol Obstet* 2002; 76: 319-320.