Hemorrhagic Corpus Luteum Cyst Torsion in Term Pregnancy: A Case Report

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Hemoperitoneum during pregnancy resulting from spontaneous rupture of adnexal torsion is a rare cause of fetal and maternal death. Presenting symptoms include severe abdominal pain, followed rapidly by maternal shock and fetal distress. It is hard to localize the adnexae in advanced pregnancy. Here, we present a case of spontaneous rupture of hemorrhagic corpus luteum cyst torsion that had not been previously diagnosed by ultrasound during term pregnancy. The patient was sent to our emergency room for sudden onset of severe low abdominal pain. Treatment consists of maintenance of adequate circulating intravascular volume and rapid surgical intervention. Preoperative diagnosis of adnexal torsion during term pregnancy is very difficult, although it is always identified during surgery.

Key Words: hemorrhagic corpus luteum, cyst torsion, term pregnancy

Adnexal torsion is a rare occurrence during pregnancy. It has been described as a severe complication of ovarian hyperstimulation syndrome and after ovarian stimulation for in vitro fertilization (IVF), with the highest incidence during the first trimester [1]. It is rare during the second trimester [2] and exceptional during the third trimester [3]. Adnexal torsion is generally diagnosed during surgery. A case of hemoperitoneum during term pregnancy resulting from rupture of hemorrhagic corpus luteum cyst torsion is presented in this report.

Case Presentation

A 35-year-old female patient, gravida 2, para 0, who had had uneventful antenatal care, presented at the emergency service with the chief complaint of acute abdominal pain and loss of fetal movement for 4 hours. There was no history of vaginal bleeding, abdominal trauma, fever, vomiting, headache, blurring of vision, or oliguria. Physical examination showed that her consciousness was clear and she was coherent and afebrile but irritable, with a pulse rate of 140/minute, blood pressure of 90/60 mmHg, and marked pallor. The uterus was of term size, tense and tender, with a well-defined contour and no fetal heart beat. Abdominal ultrasound confirmed intrauterine fetal death (IUFD) with the placenta in the upper uterine segment and no retroplacental blood clot. The adnexae and cul-de-sac were poorly identified. Laparotomy was performed immediately for acute abdominal pain with term pregnancy. At laparotomy, right adnexal torsion and internal bleeding were observed (Figure 1). Blood was found in the peritoneum (1,200 mL). Right salpingo-oophorectomy was performed and the left ovary was unwound and preserved. A lower segment cesarean section was then performed and a fresh stillborn female weighing 3,800 g was delivered. The placenta was fundal in location with no retroperitoneal blood clot and the uterus was intact. Four units of...
blood were transfused intraoperatively. The postoperative period was uneventful. The removed ovary weighed 65.1 g and measured 13 x 11 x 5.5 cm. Grossly, it was brown and coated with blood clots. It contained multiple cysts, a nodule, and diffuse hemorrhage. The fallopian tube measured 6.1 cm in length and 0.8 cm in diameter. Grossly, it was brown-black. Microscopically, there was intense hemorrhagic infarction of the ovary (Figure 2): there were multiple degenerated or destructed cysts, including multiple hemorrhagic corpus luteums and several follicular cysts. There were aggregates of inflammatory cells. The fallopian tube revealed congested vessels and focal hemorrhage (Figure 3).

**DISCUSSION**

Adnexal torsion is an uncommon cause of surgical emergency [3]. It usually occurs during the reproductive years and represents 2.7% of all gynecologic emergencies [3]. Its incidence is 1 in 5,000 during pregnancy, occurring more frequently in the first trimester after IVF and ovarian stimulation as treatment for infertility [3]. The proportion of adnexal torsion at the outset of pregnancy varies from 18% to 28.7%, according to previously reported series [4–12]. The condition is rare during the second trimester [2] and exceptional during the third trimester [3].

Adnexal torsion consists of total or partial rotation of the adnexa around its vascular axis. Clinical findings vary depending on whether the rotation is sudden and complete or progressive. With progressive torsion, lymphatic drainage is compromised, which leads to massive enlargement of the ovary due to lymphatic edema [13]. This is followed by venous obstruction and hemorrhagic infarction [14]. The final step is interruption of the arterial blood supply, which may result in gangrene, infection, peritonitis, and possible death [15]. Rupture of adnexal torsion during pregnancy may also occur secondary to softening of the lesion following stroma decidualization [16]. Therefore, the clinical presentation is variable, ranging from a benign asymptomatic state to circulatory collapse. Mild cases require observation, while, in severe cases, laparotomy is mandatory. The sonographic detection rate of adnexal masses during pregnancy is approximately 1% [17]. Although the

**Figure 1.** Both right ovary and right fallopian tube are twisted. The right ovary appears as an enlarged cystic mass with a ruptured hole (arrow). An area of hemorrhagic infarction is evident (arrowhead), as are hemoperitoneum and blood clot (white arrow).

**Figure 2.** Cystic changes in the corpus luteum with intense hemorrhagic infarction. (Hematoxylin & Eosin, original magnification, x 8)

**Figure 3.** Marked congestion and hemorrhage in the fallopian tube. (Hematoxylin & Eosin, original magnification, x 4)
mass had been followed sonographically, the growing uterus made detection of the persisting mass impossible. The management of such sonographically identified adnexal masses in the gravid patient remains controversial [18]. Torsion is seen two or three times more frequently in the right fallopian tube than in the left [19], which is consistent with our case. This is thought to be due to the prevention of torsion by the sigmoid colon on the left side or to slow venous flow on the right side, which may result in congestion [19].

This case illustrates the importance of considering adnexal torsion in the differential diagnosis of abdominal pain in term pregnancy, especially when ultrasound examination reveals no specific finding. Thus, ultrasonography is not always useful in the detection of adnexal masses, especially in term pregnant women. Since the rupture of the twisted ovarian corpus luteum cyst leads to massive internal bleeding, IUFD may be caused by acute hemodynamic changes in the mother in shock. Ultrasound examination did not disclose the right tortuous adnexal mass and hemoperitoneum because both a poorly distended bladder caused by anuria in shock and an enlarged uterus in term pregnancy attenuated the resolution of the ultrasound. Rapid diagnosis of this rare complication is essential because these patients are often first seen in the emergency service. Aggressive fluid and blood replacement together with prompt surgical intervention provide the only chance for a favorable outcome for both mother and child. Even with these interventions, fetal mortality remains high at 31% [20]. Our patient underwent definitive surgical treatment within hours of the initial presentation.

REFERENCES