Ruptured ovarian cysts and bilateral ectopic pregnancy complicating a case of severe ovarian hyperstimulation syndrome

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ABSTRACT

Ovarian hyperstimulation syndrome (OHSS), ruptured ovarian cysts and bilateral ectopic pregnancies are all well-recognized entities occurring in association with infertility treatment. We are reporting a case of severe OHSS which was complicated initially by ruptured ovarian cysts and later by bilateral ectopic pregnancy. Diagnosis of tubal pregnancy was obscured by stimulated ovaries, which prevented accurate ultrasound definition. The role of transvaginal ultrasound, serial beta human chorionic gonadotropin and the place of paracentesis in diagnosing these cases are discussed.

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Ovarian hyperstimulation syndrome (OHSS) is an iatrogenic condition with a varied spectrum of clinical and laboratory manifestations. The severe form is potentially lethal.[1,2] Tubal ectopic pregnancy is a common, life-threatening situation in gynecology that requires rapid and accurate diagnosis and treatment. However, bilateral ectopic pregnancy is rare, varying in frequency between one per 725 and one per 1,580 ectopic pregnancies.[3] We report an extremely rare case of OHSS, with bilateral ectopic gestation and ruptured ovarian cysts. These rare cases, may present a clinical dilemma. Therefore, this case has been reported to highlight these complications in the differential diagnosis when dealing with OHSS cases and to illustrate the difficulties that could be encountered in diagnosis and management of such cases.

Case Report. A 29-year-old nulliparous woman presented to the emergency room with complaints of lower abdominal pain, distension and nausea 14 days following ovulation induction (carried out in a private clinic elsewhere) by gonadotropin (Puregon, recombinant human follicle stimulating hormone). Soon after admission, she developed shortness of breath and oliguria. The diagnosis of severe OHSS was confirmed by ultrasound scan revealing enlarged, multi-cystic ovaries 11 x 12 cm each, ascites and bilateral pleural effusion. Blood investigations showed hemoconcentration (hemoglobin of 17.5 g/dl, hematocrit of 50%), low serum albumin and deranged renal function test. The circulatory volume was restored with intravenous fluid and human albumin. Abdominal and chest paracentesis was performed under ultrasound guidance as her condition deteriorated. Following that, her condition improved and she was discharged home. At that time, her serum beta human chorionic gonadotropin (ß-hCG) was 28 IU/L. She was readmitted a day later (3 weeks post [last menstrual cycle (LMP)]) with abdominal pain and vulval swelling. Her

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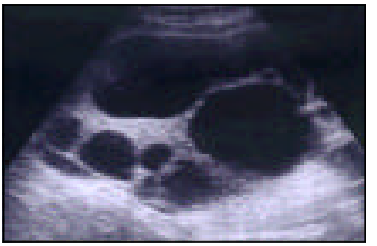

Figure 1 - Transabdominal ultrasound showing enlarged multicystic ovary.

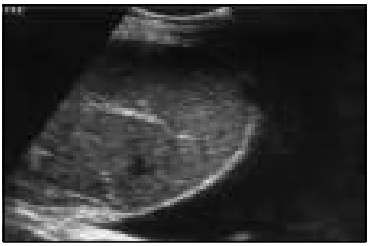

Figure 2 - Transabdominal ultrasound showing uterus with thicken endometrium and ascites.

general condition was otherwise, stable and was managed conservatively. Ten days later, while still in the hospital, she reported shortness of breath, abdominal pain and generalized weakness. She was sick looking, pale and dyspneic. Chest examination showed reduced air entry in the left lung and chest x-ray confirmed the presence of left pleural effusion. The abdomen was distended with tense ascites and tender. Trans-abdominal ultrasound showed massively enlarged, polycystic ovaries with ascites. Her hemoglobin dropped from 12.6-7.8 g/dl. Serum ß-hCG was 627 IU/l. Chest and abdominal paracentesis drained bloodstained fluid raising the possibility of ruptured ovarian cysts. Hence, she underwent exploratory laparotomy through a sub-umbilical midline incision. This showed 1500 ml of bloodstained ascites, both ovaries were polycystic and enlarged to approximately 15 x 15 cm. Two cysts in the right ovary were ruptured and bleeding. The fallopian tubes were normal in both sides. A thorough lavage was carried out, and hemostatic sutures were applied to control the bleeding. She received 3 units of blood. Postoperative recovery was uneventful. She was discharged home to be followed in the outpatient department. At that time, her serum ß-hCG was 1319 IU/L, rising to 2877 IU/L after 4 days (at 6-week post LMP). Transvaginal (TVS) and transabdominal ultrasonography showed empty uterus, thickened endometrium, bilaterally enlarged polycystic ovaries and ascites highly suggestive of ectopic gestation (**Figures 1 & 2)**. However, she declined intervention at that stage and requested to be managed conservatively as an outpatient. At 7 weeks gestation, she presented with right lower abdominal pain and per vaginal spotting. Serum ß-hCG was 4486 IU/L. Transvaginal sonography showed ascites but failed to show an intrauterine gestational sac or adnexal masses. Exploratory laparotomy was performed through the patient's prior scar. Approximately 1700 ml of fresh blood and clots were removed from the peritoneal cavity. Both ovaries were enlarged. A left ampullary tubal pregnancy 2 x 3 cm was found with active bleeding from the ostium. Hence, left salpingectomy was carried out. The right tube was distended at the ampullary region with blood clots seen at the ostium. Milking of the right tube revealed tissue consistent with product of conception. Surgical homeostasis was obtained. Histology report confirmed the presence of chorionic villi from both sites. Her postoperative course was unremarkable, and she was discharged on the 8th postoperative day.

Discussion. Ovarian hyperstimulation syndrome is an iatrogenic complication of ovulation induction therapy. The severe form is rare, with a reported incidence of 0.3-0.9%.[1] It involves massive ovarian enlargement, fluid accumulation in the peritoneal, pleural and rarely pericardial cavities, resulting in intravascular volume depletion, renal failure and hypovolemic shock. It is potentially lethal condition.[1,2] The incidence of ectopic pregnancy has remained static in recent years (11.1 per 1000 pregnancies).[4] However, it continues to be a major cause of maternal death in the first trimester.[5] The association between OHSS and ectopic pregnancy is well recognized. However, bilateral ectopic pregnancy is a rare phenomenon, varying in frequency between one per 725 and one per 1,580 ectopic pregnancies.[3] We are reporting a case of severe OHSS, which was complicated by ruptured ovarian cysts and bilateral ectopic pregnancy.

The management of OHSS is mainly supportive, while awaiting spontaneous resolution.[1] However, if pregnancy occurs, OHSS will present in a severe and protracted course as in our case, where it took nearly 50 days for the condition to resolve. Paracentesis has been proposed as an efficient way

to relieve patient's symptoms and improve cardiac and renal function. It is recommended to be carried out under ultrasound control to avoid injury to the ovaries.[6] Considering that, and the fact that hyperstimulated ovaries are liable to rupture spontaneously, we were not sure, in our case, which of these factors were responsible for rupture of the cysts.

At the initial laparotomy, both tubes felt normal. That actually emphasizes the fact that it is usually difficult to diagnose very early ectopic pregnancies. Furthermore, transvaginal ultrasonography failed to demonstrate extrauterine pregnancy. This highlights the difficulty in diagnosing ectopic pregnancy in association with OHSS. This is due to hyperstimulated ovaries obscured the accurate ultrasound diagnosis and symptoms of ectopic pregnancy may be attributed to OHSS. Moreover, the presence of ectopic pregnancy will aggravate the symptoms of OHSS. In our case, ectopic pregnancy was not visualized in spite of using high resolution TVS. This suggests that TVS may not be a reliable tool in diagnosing an ectopic pregnancy with concomitant OHSS. Therefore, serial rising ß-hCG level should alert the clinician to the diagnosis as in our case.

Early paracentesis has been reported as a diagnostic tool in suspected cases of ectopic pregnancy with severe OHSS, in whom symptoms of OHSS fail to improve with conservative therapy or become aggravated and TVS fails to diagnose intrauterine pregnancy.[7] In these reports, if ascetic fluid was blood tinged this confirmed ectopic pregnancy. In our opinion, this approach can demonstrate cases of ruptured ectopic pregnancy but will fail in cases of intact extra uterine gestation.

At the time of surgery, in order to avoid missing the possibility of bilateral ectopic gestation, a meticulous inspection of the entire pelvis is necessary. Attention focused exclusively on the bleeding left ampullary ectopic in our case, would have led to missing the intact ectopic pregnancy on the contralateral side.

In conclusion, this case emphasizes the need to maintain extrauterine pregnancy high in the differential diagnosis with the possibility of multiple ectopic pregnancies, specifically in patients on infertility treatment. One should always be aware of this diagnosis and when clinical suspicion warrants it, it may be crucial to offer laparoscopy, laparotomy, or both, to investigate this possibility, to prevent a life threatening situation for the patient.

References

1. Royal College of Obstetrician and Gynaecologists. Management and Prevention of Ovarian Hyperstimulation Syndrome (OHSS). RCOG Guideline No 2. London (UK): RCOG Press; 1995.
2. Kaiser UB. The Pathogenesis of the Ovarian Hyperstimulation syndrome. ***N Engl J Med*** 2003; 349: 729-732.
3. Rondeau J, Hibbert ML, Nelson KM. Combined Tubal and Cornual Pregnancy in a Patient Without Risk Factors a Case Report. ***J Reprod Med*** 1997; 42: 675-677.
4. Royal College of Obstetrician and Gynaecologists. The Management of Tubal Pregnancy. RCOG Guideline No 21. London (UK): RCOG Press; 2004.
5. Lewis G, Drife J, editors. Why Mothers Die 1997-1999. The Fifth Report of the Confidential Enquiries into Maternal Death in the United Kingdom. London (UK): RCOG Press; 2001.
6. Rizk B. Ovarian Hyperstimulation syndrome. In: Studd J, editor. Progress in Obstetrics and Gynaecology. Volume 11. New York (NY): Churchill Livingstone; 1994. p. 311-349.
7. Moosburger D, Tews G. Severe Ovarian Hyperstimulation syndrome and Combined Intrauterine and Tubal Pregnancy after In-Vitro Fertilization and Embryo Transfer-case report. ***Hum Reprod*** 1996; 11: 68-69.
8. Sindos M, Wang TF, Pisal N, Eben F, Singer A. Bilateral Haematosalpix in a case of ectopic pregnancy: a Clinical Dilemma. ***Am J Obstet Gynecol*** 2003: 189: 892-893.