A Case of Isolated Hemorrhagic Pleural Effusion: A Rare Presentation of Ovarian Hyperstimulation Syndrome (OHSS) – A Case Report

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Abstract- Introduction: Ovarian hyperstimulation syndrome (OHSS) is a rare, life-threatening serious complication of ovulation induction with human chorionic gonadotropin (hCG). (4) 3% of patients undergoing IVF (in vitro fertilisation) develop OHSS. But radiologically evident pleural effusions develop only in 1% among which hemorrhagic effusions are very rare (1).

Pleural effusions due to OHSS are usually associated with ascites. Isolated unilateral pleural effusions are uncommon. (2,3)

The syndrome occurs in the luteal phase or during early part of pregnancy. The syndrome was first described in 1941 and the first fatal case of OHSS with renal failure and death was described in 1951.

Keywords: ovarian hyperstimulation syndrome (OHSS) pleural effusion hemorrhagic unilateral isolated.

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Abstract: Ovarian hyperstimulation syndrome (OHSS) is a rare, life-threatening serious complication of ovulation induction with human chorionic gonadotropin (hCG). 3% of patients undergoing IVF (in vitro fertilization) develop OHSS. Pleural effusions due to OHSS are usually associated with ascites. Isolated unilateral pleural effusions are uncommon. The syndrome occurs in the luteal phase or during early part of pregnancy. The syndrome was first described in 1941 and the first fatal case of OHSS with renal failure and death was described in 1951. The number of patients who undergo infertility treatment at IVF centers has been increasing. Therefore, it should be kept in mind that there may be unilateral pleural effusion without peritoneal fluid in OHSS which can be life threatening and needs to be evaluated as soon as possible. Physicians should consider this potentially life-threatening diagnosis in all patients who undergo ovarian hyperstimulation.

Here, we present a case with ISOLATED UNILATERAL HEMORRHAGIC PLEURAL EFFUSION due to OHSS.

Keywords: ovarian hyperstimulation syndrome (OHSS) pleural effusion hemorrhagic unilateral isolated.

I. Case Report

A 30-year old nulliparous woman who has been married for 4 years with no significant medical history enrolled in an IVF center. On the second day of the menstrual cycle there were 9 antral follicles on each ovary and the levels of follicle stimulating hormone (FSH), luteinizing hormone and estradiol (E2) were 6.26 mIU/ml, 2.14 mIU/ml and 18 pg/ml respectively. The spermiogram parameters are normal {sperm count-200million/ml, percentage of motile cells-79%, percentage of abnormal cells-34%}. Ovarian stimulation was initiated with 200 IU of recombinant FSH (rFSH) for 5 days. This dose was increased to 250 IU on the fifth day because of low levels of E2 and low ovarian response. On the 10th day of induction there were two follicles that reached the size of 20 mm and 18 mm as seen on ultrasonography. Her peak E2 level was 1934 pg/ml. Next 10,000 IU urinary human chorionic gonadotropin (hCG) was injected and oocyte pickup (OPU) was performed at the 36th hour. A total of 10 oocytes were retrieved. One embryo transfer (ET) was performed on the third day of OPU. On the 12th day of ET the β-hCG level was 378 IU/ml.

On the seventh day after she was β-hCG positive she presented with complaints of dyspnea, cough and chest pain. She had tachypnea (respiratory rate-26/min) and tachycardia (pulse rate-123/min). She had a weight gain of 2 kg and she was afebrile. Her Oxygen saturation on room air was 90% as measured on pulse oximetry. There were no complaints pertaining to the abdomen (nausea, vomiting, abdominal distention).

Lab investigations revealed hematocrit of 42% and normal electrolytes, liver and renal functions. Chest x ray was not performed in view of her pregnancy and ultrasound of the abdomen revealed a moderate right pleural effusion. Her echocardiography and electrocardiography (ECG) did not revealed any abnormality. The patient was subjected to an abdominal ultrasound also and the ovaries were enlarged bilaterally (right: 86×63mm; left: 87×59 mm) with no evidence of intraperitoneal fluid. Thoracentesis was performed on the affected side and nearly 1500 cc hemorrhagic coloured fluid was recovered. The fluid analysis showed protein- 4.65g/l, LDH of 101(IU/l) thus fitting into an exudative fluid as per lights criteria.

Fluid was lymphocyte and rbc predominant (hemorrhagic) Malignancy, pulmonary embolism were considered in the differential diagnosis and pleural fluid was screened for malignant cells and bilateral lower limb venous Doppler was done which did not revealed any deep vein thrombosis.

She was followed closely as an outpatient and she recovered fully without any sequele.

II. Discussion

Although it varies depending on the level, the pleural cavity has a width of approximately 18–20 μm. The pleural membranes do not touch each other, which makes it a real gap, not a potential space. Classically, pleural effusion is the accumulation of fluid in the pleural cavity, which may be caused by any reason (5). OHSS is a rare, usually self-limiting, life-threatening iatrogenic
complication (4). In 1975, unilateral pleural effusion in OHSS was first described (6). The risk factors for OHSS are: young age, low body mass index, polycystic ovary syndrome, increased E2 levels, a previous history of the presence of OHSS, hypothyroidism and molar pregnancy (7). OHSS is classified as mild, moderate, severe or critical. Mild manifestations of OHSS are relatively common in induced cycles and include abdominal distension, mild nausea, vomiting and diarrhea (2,3). With progression of the illness pleural and pericardial effusion can be observed, which are regarded as severe OHSS (8). Our patient was classified as severe OHSS (based on the presence of severe dyspnea and moderate pleural effusion) even though she did not have intractable nausea or vomiting, oliguria, venous thrombosis. Severe OHSS has been reported in less than 2% of patients who require hospitalization. Early OHSS is correlated to ovarian response to stimulation and is an acute effect of the administration of exogenous hCG that usually occurs within 9 days after oocyte retrieval. In contrast, late OHSS occurs after the initial 10 day period, is only poorly correlated to ovarian response and is more correlated to the endogenous hCG produced by an implanting embryo (9). The main aim of the induction of ovulation is to achieve pregnancy but if pregnancy occurs OHSS tends to be more severe and may last longer. Although its pathophysiology is not known exactly, an increase in capillary permeability, fluid accumulation in a third space caused by this increase and inadequate organ perfusion are suspected. Vascular endothelial growth factor (VEGF), components of the renin–angiotensin system, prostaglandin, and cytokines such as interleukin (IL)-6 and IL-8 play a role in its etiopathogenesis. (10). Capillary permeability is reduced by 70% by the administration of VEGF antibodies, which is considered the most essential factor. Holes in the diaphragm and negative intrapleural pressure may draw fluid from the abdomen to the thoracic cavity (2). However, it is hard to explain unilateral pleural fluid. Although its pathogenesis is controversial, it is attributed to the fact that lymphatic drainage on the right side is less compared with on the left side, and holes in the diaphragm occur more often on the right side (2,10). In our patient pleural fluid was on the right side and there was no fluid in the abdomen (9). She recovered by pleural drainage and supportive therapy. In the literature pleural effusion may be exudative (as in our patient) or transudative (9). Because of our patient’s pregnancy, a chest X-ray could not be performed but ultrasound helped us to diagnose pleural effusion. In the literature there are reports about the use of ultrasound in pleural effusion (3,4,9). Also, ultrasound can detect as little as 5 mL pleural fluid (5). In our patient although there was a large amount of pleural effusion, no other significant markers of severe OHSS were present. If only the abdominal cavity is examined, pleural effusion could easily be overlooked. A good complete examination of an OHSS patient, early diagnosis, adequate pleural drainage, and then good supportive therapy make the prognosis of OHSS favourable.

### III. Conclusion

As a result, the number of cases resorting to the treatment of infertility and the number of centers where it is employed have been increasing. Although OHSS is considered as if it is a syndrome that belongs to gynecology and obstetrics clinics or IVF units, the chances of clinicians who work in the emergency service and thoracic diseases and thoracic surgery centers encountering these patients have increased. Therefore, it should be kept in mind that there may be unilateral pleural effusion without peritoneal fluid in OHSS.

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