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Continuous Drainage of a Pleural Effusion with a Pigtail Catheter Secondary to Ovarian Hyperstimulation Syndrome: A Case Report with Literature Review

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Abstract

Background: Ovarian Hyperstimulation Syndrome (OHSS) is an iatrogenic complication that may occur in hormonally induced ovarian stimulation cycles. Generally, the symptoms are self-limited and resolve spontaneously. Occasionally OHSS can become life-threatening secondary to complications such as venous thromboembolic events, electrolyte imbalance, organ dysfunction and massive third spacing. Pleural effusion, or fluid collection within the pleural cavity surrounding the lungs is one such negative consequence. Our objective was to describe an unusual case requiring the need for continuous drainage of a pleural effusion in a patient suffering from OHSS with minimal ascites.

Case presentation: We present a case of a patient undergoing treatment for infertility that presented to the ED with complaints of shortness of breath and abdominal pain. Reoccurence of pleural effusion on the left side prompted one-time thoracentesis. Reoccurrence of her left lung pleural effusion prompted a one-time left thoracocentesis, which was complicated by a pneumothorax. Eventually, the patient did well and went on to have an uncomplicated delivery.

Conclusion: Although pleural effusions with the absence of clinically significant ascites secondary to OHSS are not readily diagnosed they carry significant morbidity. In this report, early diagnosis and intervention with the use of a continuous pigtail drain resulted in an excellent patient outcome.

Keywords: Ovarian hyperstimulation syndrome; *In vitro* fertilization; Pleural effusion; Pneumothorax; Thoracocentesis

Introduction

Ovarian Hyperstimulation Syndrome (OHSS) is often noted to be the most serious complication of assisted reproduction [1] occurring in 0.3-5% of women undergoing assisted reproductive cycles with ovarian stimulation [2]. Although the pathophysiology of OHSS is not completely understood it is thought to represent an over expression of normal ovulatory processes caused by inflammatory mediators most notably Vascular Endothelial Growth Factor (VEGF) deranging vascular permeability [3,4]. This causes a fluid shift from the intravascular space to the interstitial or third space, and this process is associated with the complications of the syndrome [5]. It is considered to be an iatrogenic disorder affecting generally healthy women [5] and carries significant morbidity and mortality.

Despite strategies to decrease the incidence of OHSS including close observation, adherence to strict guidelines limiting the rise of estradiol during stimulation, using GNRH agonist triggers for ovulation, and criteria for cancelling cycles, there remains a potential risk for OHSS with ovarian stimulation. We describe a case of OHSS which was complicated by bilateral symptomatic pleural effusions treated by continuous pigtail catheter drainage.

Case Report

A 30-year-old female underwent ovarian stimulation using a "down regulation" protocol with GnRH antagonist. She subsequently underwent intracytoplasmic sperm injection secondary to male factor infertility. On day 10 of the cycle, she was given 5000 IU of hCG and her estradiol level was 4029 pg/mL. Oocyte retrieval was performed and she had Embryo Transfer (ET) with 2 fresh embryos.

Two days following embryo transfer the patient presented to the emergency department with worsening abdominal discomfort and distention with associated shortness of breath. Physical exam was remarkable for generalized periumbilical tenderness to palpation. Her lungs were clear to auscultation at apices, with diminished breath sounds noted in the lower lobes and dullness to percussion in right lower lobe.

Laboratory studies obtained revealed no electrolyte abnormalities, hemoglobin of 13.4 gm/dL and serum hCG of 730 mIU/mL. Liver and renal function tests were within normal limits. The patient had adequate urine output. Urinalysis was significant for trace ketones, but was otherwise unremarkable.

Transvaginal ultrasound showed markedly enlarged ovaries with multiple anechoic structures compatible with corpora lutea. Minimal to moderate amounts of free fluid was also present in the abdomen and pelvis. Additionally, a right pleural effusion was noted.

On hospital 2 ultrasound guided thoracocentesis was performed with pigtail catheter placement for treatment of her right pleural effusion. Initially, 1300 mL of fluid was drained and fluid analysis was consistent with a transudative effusion. Repeat radiologic studies were significant for a 100 mL right pleural effusion, and a 700 mL left pleural effusion. The patient remained asymptomatic and no further intervention was required. The pigtail catheter was left *in situ* for 4 days. The pigtail catheter was removed on hospital day 5 and she was discharged home the same day.

The patient was readmitted to the hospital ten days later, when

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she presented with complaints of left sided pleuritic chest pain. The patient had remained asymptomatic until two days prior to admission however physical exam findings upon readmission suggested re-accumulation of her left pleural effusion. Therapeutic thoracocentesis was performed with drainage of moderate amount of fluid and was complicated by small pneumothorax, which did not require additional treatment. Pleural effusions did not re-accumulate and the patient was discharged from the hospital the next day in stable condition. At the time of discharge she was noted to be 6 weeks and 5 days pregnant with twin gestation. The remainder of her pregnancy was uneventful and she carried the pregnancy to term.

Discussion

Ovarian hyperstimulation syndrome is believed to be an iatrogenic, but potentially serious condition, with an incidence of around 8% of stimulated ovarian cycles [6,7]. Although, generally self-limited, OHSS has the potential to be a serious complication with significant morbidity and mortality. The syndrome typically occurs during ovarian stimulation with gonadotropins with concomitant use of hCG [8,9]. The pathophysiology of the syndrome is not completely understood however several risk factors for OHSS have been identified including: younger age, the presence of polycystic ovarian syndrome, >13 follicles, rapidly rising, or serum concentration of estradiol >2,500 pg/ml, and lower FSH levels and female factor infertility [8].

Ovarian hyperstimulation syndrome can be classified into categories including mild, moderate and severe forms. The mild form presents with abdominal distension and discomfort and be can further classified into grade 1 and 2. Grade 1 is composed of chemical hyperstimulation with estrogen and pregnanediol excretion greater than 150 mg/24 hours and 10 mg/24 hours, respectively. Grade 2 is noted to have ovarian enlargement in addition to elevated estrogen and pregnanediol excretion. Progression of OHSS from the mild to moderate form includes the same constellation of symptoms and laboratory findings of the mild form with the addition of abdominal ascites noted on imaging studies. Moderate OHSS can be further subcategorized into grades 3 and 4. Grade 3 includes the characteristics of grade 2 with the addition of abdominal distension and pain, with ovarian enlargement reaching up 12 cm. Grade 4 includes the addition of systemic symptoms such as nausea, vomiting, and/or diarrhea. Further progression to the severe form includes all aforementioned signs and symptoms with significant extravasation of fluid in the form of clinically apparent ascites, pleural or pericardial effusions. Severe forms of OHSS are divided into grades 5 and 6 with ascites and/or hydrothorax characteristic of grade 5 and hemoconcentration in 6 [10]. In critical forms of OHSS, patients begin to display evidence of hypovolemic shock, as well as acute renal and respiratory failure [11]. Severe, late onset OHSS generally occurs 12-17 days following administration of hCG with establishment of pregnancy [12].

The exact pathophysiology of OHSS is not completely identified however, it is believed to be a result of overproduction of vasoactive substances from the ovary which are essential for follicle release and neovascularization of the corpus luteum [13]. Several mediators and cytokines, have been identified in the pathophysiology of OHSS such as IL-1, IL-6, IL-8, Tumor Necrosis Factor- alpha (TNF-a), and Vascular Endothelial Growth Factor (VGEF) [11,14,15] as well as hCG as been known to involved in the pathogenesis of OHSS leading to increased angiogenesis and vascular permeability secondary to its up regulation of VEGF and activation of the intra-ovarian renin angiotensin system [12]. Gonadotropic activation of these cytokines and vasoactive substances results in increased vascular permeability and resultant third spacing of fluid and the development of ascites, pleural effusions, and hemoconcentration [11]. Pleural effusions of the right lung are more common [16]. The explanation behind this is less lymphatic drainage and the diaphragmatic hollows are greater on the right side [17]. Anatomical defects in the diaphragm which allow the ascites to pass through to enter the pleural space has also been described in the literature as a contributing factor to the increased incidence of right sided pleural effusions [16,18,19].

The case we report here is noteworthy for a number of reasons. First, when severe forms of OHSS manifest, it is commonplace that they present with ascites; in our case however, ascites was not clinically evident and fluid accumulation was noted only in the chest cavity. Therefore, importantly, providers should not assume that OHSS is not severe in the absence of ascites. Secondly, this case is depicts successful management of the pleural effusion with percutaneous pigtail drainage and a repeat accumulation of pleural fluid, albeit on the other side. Although conventional treatment with thoracocentesis is a safe and efficient treatment modality and may be repeated as often as needed [20], each repetition of the procedure increases risks for patients. If the effusion does not resolve with the initial thoracocentesis repeat procedure will subject the patient to the additional discomfort and they will also endure the potential risks associated with the invasive procedure such as lung injury, bleeding, infection, and a pneumothorax.

Once the pigtail drain is placed, it will continue to help resolve the effusion, these findings are in concert with those seen by other groups [21]. In comparison of pigtail drains to conventional stiff drains they were found to have better clinical outcomes and decreased period of drain *in situ* [22]. The pigtail catheter has a considerably smaller bore then the conventional chest drain, is much less traumatic to insert, and results in a noticeably smaller residual scar [22]. Furthermore, continuous drainage catheters have been commonly used for drainage of abdominal fluid collection in more severe forms of OHSS as an alternative to repeat paracentesis [21]. Our case highlights the utility and success of pigtail catheter placement for the drainage of pleural effusion secondary to OHSS.

In conclusion, ovarian hyperstimulation syndrome can be a potentially life threatening condition if it is not promptly recognized and appropriately managed. Providers need to be vigilant of cases of OHSS, which may present with third spacing isolated to the chest cavity with the absence of clinically significant ascites. The mainstay of management of OHSS is supportive care, however, when present in a severe form, more invasive management and intervention may be necessary [22]. Furthermore, in cases where the patient presents with symptomatic pleural effusions regardless of the severity of the OHSS, a small pigtail drain may be placed for continuous drainage, which may alleviate the patient's symptoms, decrease the need for repeat thoracentesis all the while allowing the syndrome to slowly resolve over time.

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