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RESEARCH ARTICLE

OVARIAN HYPERSTIMULATION SYNDROME (OHSS) FOLLOWING A NATURAL CONCEPTION, A CASE REPORT AND REVIEW OF LITERATURE

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ABSTRACT

Introduction: OHSS is a rare complication occurs following the use of assisted reproductive technologies. Most of the current literature describes spontaneous OHSS in patients with endocrine disorders, pregnancy with triploidy & invasive mole etc. But we describe an interesting case of spontaneous OHSS with its management, which mimics as ovarian malignancy in some aspect with no risk factor to develop OHSS.

Case report: A 25yr old G4P0120 at 9+3 weeks was referred to us with severe abdominal pain & distension for 1 day. On examination she had gross ascites with bilateral ovarian cyst of size 11×7 cm & 12×9 cm. Her CA 125, βhCG, Hb&Hct were 445U/ml, 212844.26 miu/ml, 15.5 g/dl & 48% respectively. Her ascitic fluid cytology and culture for tuberculosis were normal. She was managed conservatively for 2 days. But with deteriorating general condition decision for termination of pregnancy was taken. After that she has improved both clinically, radiologically and biochemically.

Conclusion: Spontaneous OHSS is a rare condition. It should be diagnosed early. The main stay of management is conservative with proper vitals monitoring. In rare case sometime termination of pregnancy or surgical management is required to improve maternal condition.

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INTRODUCTION

Ovarian Hyperstimulation Syndrome (OHSS) is a rare iatrogenic complication of ovarian stimulation that occurs either during the luteal phase or in early pregnancy (Delvigne *et al* 2002.). Severe or critical form of OHSS is a rare complication occurs following exogenous ovarian stimulation. Most of the current literature describes spontaneous OHSS in patients with endocrine disorders, pregnancy with triploidy& invasive mole etc. The clinical manifestation varies from mild haemodynamic changes to severe life threatening conditions. But management of OHSS is same whether it is due to above listed causes or it is due to spontaneous in origin. Spontaneous form of OHSS is very rare. Though it's a rare entity, its early diagnosis is important particularly in spontaneous variety as it can mimic ovarian malignancy in some aspect with a different management protocol. Amarin ZO in 2003 described a case report where intractable OHSS was managed with laparotomy and bilateral oophorectomy. We know bilateral oophorectomy is the last option one can think of in the management of OHSS and it is not recommended for the same (Bellver *et al*., 2003).

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We describe an interesting case of spontaneous OHSS which mimics as ovarian malignancy in some aspect, where apparently no cause was identified and was managed with termination of pregnancy with subsequent improvement in general condition of the patient.

Case report: A 25yr old G4P0120 at 9+3 weeks was referred to us with severe abdominal pain for 1 day. Her obstetric history revealed one fresh still birth at 8th month of gestation and two second trimester abortions for unwanted pregnancy. Her previous pregnancies were inadequately supervised. This pregnancy was a spontaneous conception. She did not report any intake of medication before or during her present pregnancy except folic acid. Her abdominal examination revealed tense ascites with generalized tenderness. A high resolution USG was performed, showing bilateral multi cystic hypoechoic ovarian masses of size 11 × 7cm and 12 × 9 cm, with no solid area or septation with gross ascites. Her obstetrics scan revealed a single live intrauterine pregnancy corresponding to 8+6 weeks which also rules out any possibility of a molar pregnancy. Her βhCG value was 212844.26 mIU/ml. Thyroid function tests were normal. Her CA 125 was 445 U/ml. X ray chest revealed no abnormality. Haemoglobin and haematocrit values were 15.5 g/dl, and 48% respectively.

Laboratory examinations showed normal platelet, liver & renal function tests. Patient was managed conservatively. CVP (central venous pressure) catheter was inserted. Emphasis was given for the proper maintenance of hydration throughout her hospital stay. Prophylactic unfractionated heparin 5000 IU s.c. twice a day was started. Over the period of 48 hours her liver function deranged to more than two time of normal value with increasing ascites and later on she developed oliguria. Her serum albumin was 2.3 mg/dl. Though we have kept spontaneous OHSS as the provisional diagnosis, because of its rarity of presentation we did investigation to rule out pregnancy with tuberculosis ormalignancy. Paracentesis was done twice to relieveabdominal distension. Fluid was also sent for malignant cytology andwas examined for any evidence of abdominal koch's and found to be negative for both. An informed consent for the termination of pregnancy was taken in view of deteriorating maternal condition with severe OHSS. Pregnancy was terminated by suction and evacuation at 9+6 weeks, tissue sent for histological examination showed normal product of conceptions.

A repeat βhCG 7 days after termination of pregnancy was 3178 mIU/ml. Patient improved after the pregnancy was terminated. A repeat USG was performed after 45 days of abortion showed normal size ovary.

DISCUSSION

Spontaneous OHSS is an extremely rare pregnancy event. Assisted reproductive technologies are commonly associated with OHSS. Most of the case reports published in literature on spontaneous OHSS was because of hypothyroidism (Ilanchezhian *et al*., 2015), hyperthyroidism (Marciniak *et al*., 2009), triploidy (Rachad *et al*., 2011) or multiple pregnancy (Agrawal *et al*., 2012). But we describe a case where no apparent cause is identified which showed more research is needed on this particular area. The basic mechanism behind OHSS in most of the patients is due to excess of hCG. There is a direct relationship between the hypothyroidism and OHSS. Studies claimed that high levels of thyroid stimulating hormone can stimulate ovaries and can leads to the development of OHSS, although we didn't found it in our case. Spontaneous forms of OHSS generally develop between 8 and 14 weeks of POG (period of gestation), differing from iatrogenic OHSS, which usually starts between 3 to 5 weeks of pregnancy. In our patients we diagnosed it at 9+2 weeks of gestation. Pregnancies with multiple gestation and invasive mole lead to increase in hCG level and may result in spontaneous OHSS.

We have excluded both of these conditions by confirming a normal single intrauterine gestation on USG and by histopathological examination of products of conception. Although the aetiology still remains unclear, various theories have been proposed to define the pathophysiology behind OHSS. Whatever may be the inciting event, all leads to a final event of increased vascular permeability and may lead to the development of ascites and pleural effusion. Earlier case reports by (Lipitz *et al*., 1996) managed such type of clinical scenario with laparotomy suspecting ovarian malignancy. As OHSS mimics ovarian malignancy in some aspect. It should be considered as a differential diagnosis while managing pregnancy with ovarian cyst. In our patients we also had some features suggestive of ovarian malignancy like sudden onset of spontaneous ascites, enlarged ovary with deteriorating maternal condition & raised CA 125. But these parameters should be carefully interpreted as it may be elevated in patients with OHSS. Increase in CA 125 concentration in OHSS occurs probably because of increased mesothelial expression of the antigen. For such clinical scenario, conservative versus laparoscopic management of ovarian cyst should be carefully made. Recent case reports on OHSS suggest that in some patient with mutated FSH receptor can lead to recurrent OHSS (Meduri *et al* 2008, Dieterich *et al* 2010). These patients with mutated FSH receptor usually presented with recurrent OHSS during pregnancy. As in this case she didn't have any previous history of spontaneous OHSS we didn't investigate for such mutation in our patient. In our case she didn't have any risk factor for the development of OHSS. She recovered both clinically, biochemically and radiologically after termination of pregnancy. Rosen & Lew *et al* (1991), Zalel *et al* (1992) Bibishahnaz *et al* (2008) reported three separate cases where pregnancies were terminated in view of severe grade III OHSS as we did in our case. But there were various other case reports by Todros *et al* (1999), Shergill KS (2005) & CepniI *et al* (2006) where conservative management followed a delivery of a healthy baby but the risk of continuing pregnancy always be weighed against the deterioration of the maternal condition. The management of OHSS should be tailored to the degree of severity, but it requires careful monitoring. Most of the mild to moderate OHSS patients can be managed as outpatient basis except patient with severe and critical OHSS need admission.

They should be closely monitored to ensure that they do not progress into the critical category. In patients with significant ascites, paracentesis is helpful, by decreasing intra-abdominal pressure and improving renal blood flow with a subsequent increased production of urine. In literature there are very few cases where termination of pregnancy was required though it should be the last thing to be done. It may be an option for patients with life threatening situation where she is not responding to the conservative management. Surgery should be considered in cases with torsion, ovarian rupture or in cases of ectopic pregnancy by an experienced surgeon (RCOG green top guideline no 5, 2016). Conclusion- Our case emphasizes the possibility of OHSS in spontaneous pregnancy without any known risk factor. When OHSS concomitant with spontaneous pregnancy is diagnosed, continuation of the pregnancy should be encouraged, but it may not be possible always as in our case. Main stay of management of OHSS is conservative with emphasis on fluid management, Prophylactic anticoagulation and strict vitals monitoring. One has to be more vigilant for any fatal complications as even death has been reported in the literature. Surgical intervention should be avoided for OHSS by careful evaluation of the patient. In this case We didn't find any cause of OHSS for her, may be further research on the pathophysiology in future will reveal insight to the development of OHSS.

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