INTRODUCTION

Very rarely Ovarian Hyperstimulation Syndrome (OHSS) has been described in normal spontaneous pregnancy. Ovarian Hyperstimulation Syndrome (OHSS) is a rare, iatrogenic complication of ovarian stimulation with follicle stimulating hormone (FSH) medications. OHSS was first described in 1943, and the first fatal case was documented in 1951. It usually occurs in patients with polycystic ovarian syndrome, in whom ovulation induction is done by gonadotrophins. The incidence of PCOS in general population is 20-33 %. Severe OHSS is estimated to occur in approximately 1% of all gonadotropin cycles. Hyperstimulated ovaries release a number of vasoactive mediators under the influence of hCG. These include vascular endothelial growth factor (VEGF) and several pro-inflammatory cytokines that interact to produce the characteristic pathophysiology of OHSS. This is marked by increased capillary permeability, leakage of fluid from the vasculature, third space fluid accumulation and intravascular dehydration.

Its clinical manifestations are ovarian enlargement, ascites, oliguria, abdominal pain, electrolyte imbalance, hemoconcentration, and even thrombosis in severe cases. In spontaneous pregnancy, OHSS is an extremely rare event. Here we present a moderate OHSS case, in a spontaneously conceived twin pregnancy.

CASE REPORT

A 24 years old lady, referred to us with complain of amenorrhea, nausea, vomiting, lower abdominal pain and vaginal spotting. She was in her second ongoing pregnancy; her previous obstetrical history was unremarkable. She had one year old baby girl.

Her menarche was at the age of 13 years and subsequent menses were regular. She was amenorrheic for last 10 weeks. She had no history of infertility, ovulation induction or any other medication in the last one year.

There was no history of twin pregnancy and polycystic ovaries in the family. Physical
examination revealed normal vital signs, abdominal distention and tenderness in lower abdomen. No palpable mass in abdomen. Laboratory test revealed normal Hb, hematocrit, serum electrolytes, creatinin, blood urea nitrogen and LFT’s. Abdominal USG revealed an intrauterine twin pregnancy with bilateral ovarian enlargement. Rt ovary was 12×12cm, left ovary 14×12cm in size. Moderate amount of fluid was seen in peritoneal cavity. Diagnosis of moderate spontaneous ovarian hyperstimulation with twin pregnancy and threatened miscarriage was made. Pt was admitted and managed conservatively with complete bed rest, high protein diet and intra venous fluids. After two days her bleeding and pain settled and she was discharged after explaining the condition. Two weeks later she was admitted again with same complaint. On USG the ovaries were of the same size. She got admitted 4 times till 22 weeks of pregnancy with complain of lower abdominal pain and bleeding p/v. At 24 weeks of gestation the size of her ovaries started decreasing and reached near normal till 26 weeks of gestation. At 27 weeks she had PPROM for which she was referred to tertiary care hospital for good neonatal care, after that she lost her follow up but came again at 33 weeks with labour pains. Her LSCS was done due to fetal distress of 2nd twin. At operation her both ovaries were of normal size and shape. Two babies (both boys) delivered with first cephalic with intact membranes and second breech with ruptured membranes. Both were of 1.9 kg, remained in NICU for 3 weeks and discharged healthy.

DISCUSSION

OHSS has life threatening complications such as venous and arterial thromboembolism. Indeed, Schenker and Ezra reported the death of patients with OHSS to be caused by these complications consequently, one must be aware of the rare but possible occurrence of OHSS in spontaneous pregnancy, in order to prevent its complications. Ovarian hyperstimulation syndrome in spontaneous pregnancy is an extremely rare event. Under certain circumstances such as twin pregnancies, the possibility of its existence may be higher because of higher HCG concentrations during the early pregnancy.

OHSS in Spontaneous pregnancy usually develop between 8 and 14 weeks of amenorrhea, differing from iatrogenic OHSS, which usually starts between 3 and 5 weeks of amenorrhea. The recent identification of mutations in the follicle stimulating hormone (FSH) receptor gene, which display an increased sensitivity to HCG and are responsible for the development of spontaneous OHSS, helps us to understand this problem. In iatrogenic OHSS, the follicular recruitment and enlargement occur during the administration of exogenous FSH. In the spontaneous form however, the follicular recruitment and growth occur later through the promiscuous stimulation, by pregnancy-derived HCG, of a mutated FSH receptor that is abnormally sensitive to HCG or a wild type FSH receptor in the presence of abnormally high levels of HCG. Thus, the symptomatology of spontaneous cases of OHSS usually develops at 8 weeks’ amenorrhea and culminates at the end of the first trimester of pregnancy.

In the literature, different cases were reported in which spontaneous pregnancy with OHSS and hypothyroidism was found together. It was claimed that high levels of thyroid stimulating hormone can stimulate ovaries in women with hypothyroidism and can cause ovarian hyperstimulation. In our case, hypothyroidism was not present.

In literature, ovarian hyperstimulation is also reported in singleton pregnancy with a mole. But in our case there was no associated molar pregnancy.

The management of OHSS is tailored to the degree of severity. Early recognition and prompt appropriate treatment will avoid serious sequelae. Severe OHSS requires hospital admission and prompt management to replace lost intravascular volume and prevent its potentially fatal complications namely renal failure and thromboembolic events. These patients should be closely monitored to ensure that they
should 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. Drainage of ascetic fluid in OHSS may be carried out abdominally or vaginally. The vaginal route has the benefit of easier access and avoidance of ovarian trauma. Pleural effusions are not uncommon in OHSS. Thoracocentesis may be necessary to avoid respiratory distress, accompanied by paracentesis in order to prevent fluid from leaking back into the pleural cavity.

CONCLUSIONS
This case report emphasizes the importance of thorough evaluation of all women presented with acute abdomen pain and ovarian masses during pregnancy. Although the condition is extremely rare, it is a potentially fatal in its severe form if not timely diagnosed and managed conservatively. With the increasing awareness of these conditions, more and more cases could be detected and reported.

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REFERENCES