A case of recurrent Spontaneous Ovarian Hyperstimulation Syndrome (OHSS): two vastly different clinical courses in consecutive pregnancies.

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Transformation: Making Waves

Background

Spontaneous OHSS is rare. There is a reported incidence of 0.5-5%. It presents at 8-12 weeks gestation, as opposed to 3-5 weeks common with assisted reproductive technologies[1].

Risk factors include young age, low BMI, PCOS, hypothyriodism, previous history of OHSS, hyatidiform molar pregnancy, multiple pregnancy and gonadotrophin-producing pituitary adenomas[2,3]. De Leener classification of OHSS divides spontaneous OHSS syndrome cases based on clinical presentation and FSH receptor mutation into four types[4]. Five mutations of the FSH receptor have been identified and OHSS is thought to occur due to abnormal sensitivity of hCG to mutant FSH receptor (FSHR) types or excessive secretion of glycoprotein in the same subunit[4].

Case Study

This case discusses a 30 year-old (G3P1) with a history of multi-fibroid uterus and endometriosis. Her obstetric history including an uncomplicated pregnancy with an emergency caesarean delivery and a miscarriage.

Following natural conception in February 2021, the woman presented to the Emergency Department at 10/40 with worsening nausea/vomiting managed with antiemetics. An ultrasound at 11+3/40 showed CRL 44mm (appropriate for dates), multiple fibroids and bilateral enlarged multicystic ovaries (Rt 724cc, Lt 870cc) with free fluid in adnexa (Rt 77cc, Lt 30cc).

The woman was symptomatic, with significant abdominal distension and pain. CA-125, serum sex hormones (oestrogen 65000, testosterone 23mmol) and bHCG (498400) were markedly elevated. Multi-D discussion suggested low risk of malignancy. Medical termination of pregnancy (MTOP) was offered at 12/40 given severity of symptoms and concern for a differential diagnosis. Following MTOP, symptoms worsened, and ovaries further enlarged on ultrasound; at largest the right Ovary measured 264x100x235mm (3244cc) and left ovary 181x128x140mm (1696cc). She required paracentesis for ascites. Histopathology confirmed normal pregnancy tissue. An MRI Pituitary was performed showing a 4mm microadenoma and a cystic structure within the anteromedial left medial cranial fossa with benign appearances. The pituitary lesion was thought to be an incidental finding and unrelated to the spontaneous OHSS presentation.

She was monitored in Clinic with weekly FSH, LH, Oestradiol and bHCG to confirm return of regular menses.

The patient was counselled on the risk of recurrence in future pregnancies.

She spontaneously fell pregnant again in November 2021 and was closely monitored throughout with serial ultrasonography. The right ovary measured 31cc and left 54cc in early pregnancy. There was recurrence of OHSS with peak enlargement at 13/40 (right ovary 100x99x99mm (545cc) and left ovary 137x95x100mm (689cc)) and only trace free fluid. The condition resolved spontaneously with only mild symptoms. The pregnancy continued uneventfully to term.

A

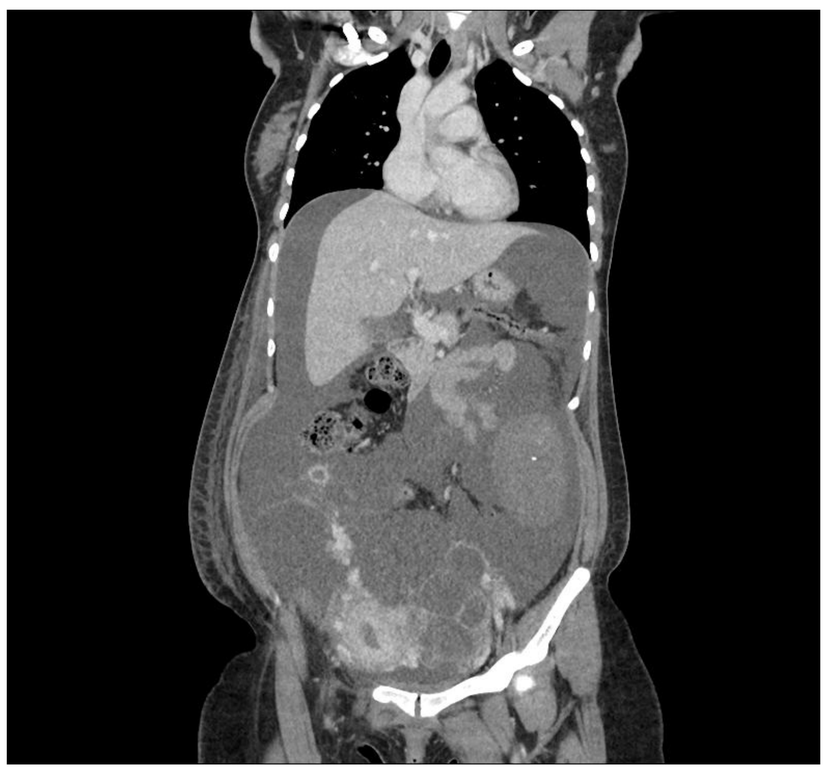
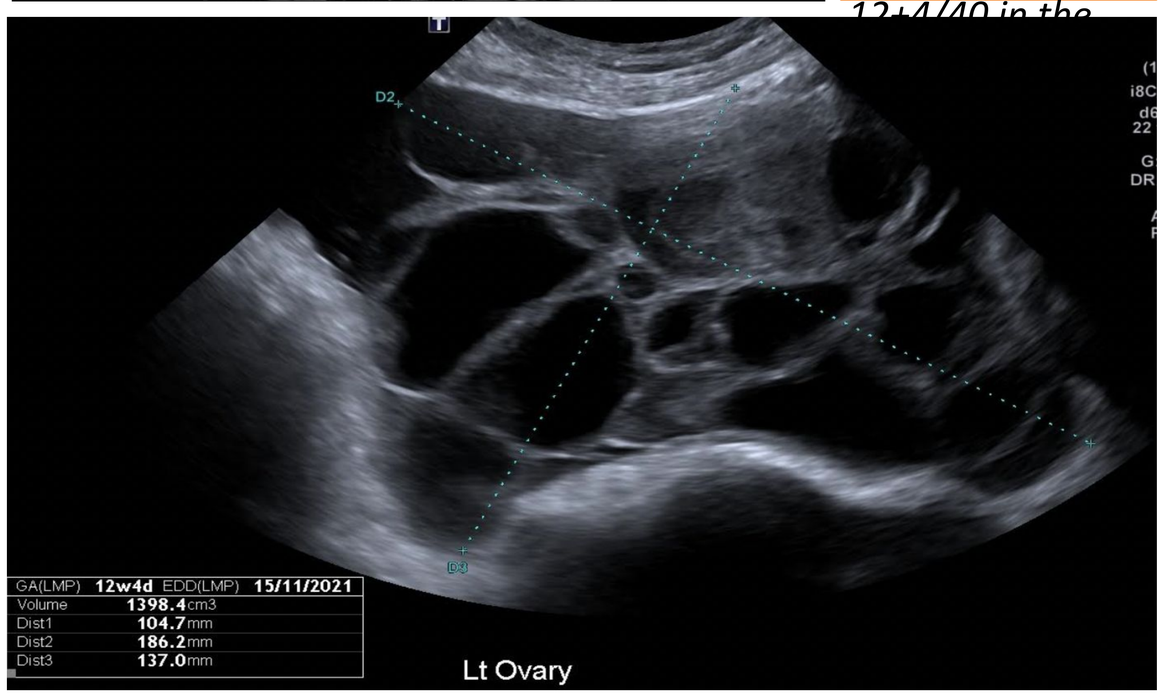


Figure A: *CT Abdomen/Pelvis demonstrating large volume abdominal ascites, enlarged multi-cystic ovaries and multi-fibroid uterus*

Figure B: *Enlarged, multi-cystic appearance of Left Ovary at 12+4/40 in the*

B



Discussion & Conclusion

Spontaneous OHSS is extremely rare. It is difficult to diagnose and presents similarly to ovarian malignancy with abdominal distension, ascites, and bilateral ovarian enlargement. Genome sequencing of FSH receptors could be undertaken to further investigate a cause for this case. Possibly, termination could have been avoided if symptoms had improved with management of OHSS, as most cases in literature have progressed to term deliveries[1].

Previous occurrence was an identifiable risk in the subsequent pregnancy. This pregnancy was closely monitored with serial ultrasonography allowing earlier detection of recurrence. Interestingly a significantly different clinical course was noted.

References

1. Sridev S, Barathan S. Case report on spontaneous ovarian hyperstimulation syndrome following natural conception associated with primary hypothyroidism. J Hum Reprod Sci. 2013 Apr;6(2):158-61. doi: 10.4103/0974-1208.117164. PMID: 24082659; PMCID: PMC3778607.

2. . Kasum M, Oreskovic S, Jezek D. Spontaneous ovarian hyperstimulation syndrome. Coll Antropol. 2013 Jun;37(2):653-6. PMID: 23941020.

3. Ayhan A, Tuncer ZS, Aksu AT. Ovarian hyperstimulation syndrome associated with spontaneous pregnancy. Hum Reprod 1996;11:1600-1

4. A. De Leener, L. Montanelli, J. Van Durme, et al. Presence and Absence of Follicle-Stimulating Hormone Receptor Mutations Provide Some Insights into Spontaneous Ovarian Hyperstimulation Syndrome Physiopathology, *The Journal of Clinical Endocrinology & Metabolism*, Volume 91, Issue 2, 1 February 2006, Pages 555–562, https://doi.org/10.1210/jc.2005-1580