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Coexisting complete mole and live fetus in DCDA pregnancy with secondary ovarian hyperstimulation and torsion: A unique situation

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Background

The risk of developing a complete hydatidiform mole with coexistent foetus (CHMCF) is estimated to be between 1:22,000-1:100,000 pregnancies. Available data suggests that there is a 21% chance of a live birth in CHMCF(1). The presence of a concurrent foetus does not appear to increase the risk of gestational trophoblastic neoplasia. While a medical termination is the usual recommendation, management is largely guided by patient wishes. Pre-eclampsia and hyperthyroidism are two of the better studied complications in CHMCF pregnancies (1). Ovarian Hyperstimulation Syndrome (OHSS) is often an iatrogenic condition associated with ovulation induction. The very rare spontaneous form is known to occur in multiple gestations and molar pregnancies, thought secondary to the high serum HCG levels(2). Only a handful of CHMCF cases with subsequent sOHSS have been documented worldwide.

Case

34 year-old G3P2 (2 term vaginal births) at 9 weeks gestation (spontaneous conception)

History: Mild vaginal bleeding, severe nausea, vomiting and generalised pelvic pain for one week

Exam: Abdomen soft, epigastric tenderness, fundal height 16 weeks
Speculum: Cervix closed, old blood in vault
Serum HCG: **>999,999**
Electrolytes: Within normal limits
FBC: Hb 83 (anaemia requiring iron infusion)
Management

- Referred for further imaging and discussion with Gynaecological Oncology
- Transferred to tertiary centre on readmission for nausea and vomiting
- Suction curette under ultrasound guidance, EBL 700 mL (uterotonics needed)

Post operative complications

- **Early onset Pre-eclampsia** requiring MgSO4 infusion and Captopril BD
- **Thyrotoxicosis** (TSH<0.05/ft4 53/antibody negative) causing tachycardia and tachypnoea (CXR clear), treated using Prednisolone and Propranolol QID

Cytogenetics: Female foetus and complete hydatidiform mole, no evidence of malignancy

Represented to local hospital 3 weeks curette with acute abdominal pain in the context of bilateral enlarged cystic ovaries and right ovarian torsion on ultrasound

- Unable to proceed with surgery due to hypertensive crisis intra-operatively
- BP stabilisation in CCU
- Transferred again to tertiary centre where laparoscopic decompression of bilateral cysts and right ovary de-torsion was performed (EBL minimal, **Right ovary preserved**)

Follow up

- HCG levels were followed weekly until negative thrice, then monthly levels for six months

Images from case

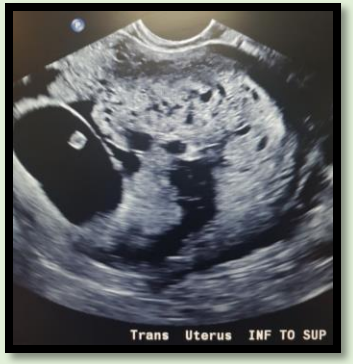

Image 1: Initial presentation scan showed hydatidiform mole encroaching on adjacent live intrauterine pregnancy

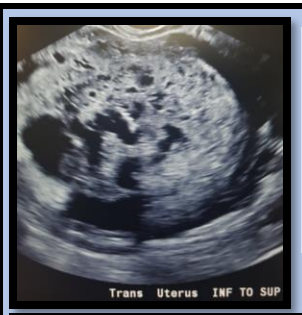

Image 2: TVUS demonstrated textbook 'snowstorm' appearance associated with molar pregnancies

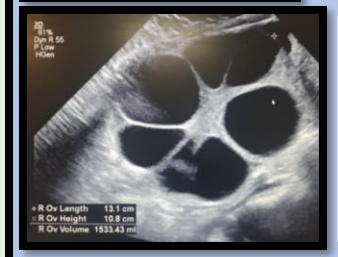

Image 3: TVUS when readmitted with sOHSS showing enlarged polycystic right ovary

Discussion

A review of the literature revealed that suction evacuation of the affected molar pregnancy appears to bring about an acute exacerbation of sOHSS (3). This usually manifests as enlarged multi-follicular ovaries and intravascular depletion from fluid loss into the third space. Possible sequelae include ascites, pericardial or pleural effusions and thromboembolism from hemoconcentration(4).

The pathophysiology of this phenomenon is unclear, but it has been hypothesised that HCG acts as a trigger for other vasoactive substances with a longer half-life (2). **A sudden decrease in HCG post evacuation has been associated with an increase in the concentration of vascular endothelial growth factor (VEGF) (5). VEGF promotes vascular permeability; this could explain the delay in onset of sOHSS.** A link to the renin-angiotensin pathway is believed to be another contributing factor to the extravasation of fluid in OHSS (2,5).

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