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"Oh My Goodness! She's got Cancer?

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Background

Hyperreactio Luteinalis is a rare and benign condition that can mimic serious malignant pathologies. Understanding the condition and optimal management is imperative for appropriately counselling patients as well as avoiding unnecessary surgical intervention. Whilst typically the disease is associated with gestational trophoblastic disease, it can rarely been unexpectedly found in normal pregnancies. In this case we discuss the incidental finding and management of hyperreactio luteinalis in a twin pregnancy at caesarean.

References

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Case Presentation

A 27yo indigenous gravid 6 para 5 female presented to a tertiary obstetric department for routine antenatal care after a dating scan had detected a dichorionic diamniotic twin pregnancy. She had a BMI of 46 and an early OGTT had confirmed the presence of gestational diabetes, however based on a HbA1c of 8.1%, it was thought more likely to be undiagnosed type 2 diabetes melllitis. Her baseline blood profile was unremarkable. High HCG levels were noted by the GP and presumed secondary to the multiple pregnancy. On the background of a previous 3b tear in P5, she indicated she would want an elective caesarean and bilateral tubal ligation. She was seen regularly throughout her pregnancy by the maternal medicine team and had several third trimester growth ultrasounds. Sonographers noted appropriate growth, but on several occasions mentioned images were limited by maternal habitus.

She was seen by the endocrinology team as management of hyperglycaemia became difficult and on general examination was noted to have features of hirsutism. The team raised the possibility of PCOS with plans for further investigation in the post-partum period.

At 35+1 weeks gestation she presenting to the delivery suite complaining of vaginal loss and contractions. She was seen by the registrar and confirmed to have PPROM as well as a 2cm dilated cervix. On a bedside US the presenting twin was breech and the decision was made for an emergency caesarean. The procedure commenced uneventfully, however at the time of tubal ligation, bilateral large multicystic, complex masses were identified measuring 10 and 9cm. Due to concern for malignancy a second consultant obstetrician was called and the joint decision for a unilateral oophorectomy was made. This was completed with no complications and sent for histopathology. The procedure was otherwise unremarkable with an EBL of 900ml. The patients post-partum recovered was uneventful. She was seen for a review of results 6 weeks post where a diagnosis of Hyperreactio Luteinalis was made based on findings.

Discussion

Hyperreactio luteinalis is a rare condition typified by bilateral cystic ovarian enlargement. It was first described by Burger in 1938 and since then 96 cases have been published. The benign finding is secondary to exaggerated physiological response to follicle stimulation. For this reason, they are most often noted in association with a number of conditions with high circulating gonadotrophin levels including gestational trophoblastic disease, polycystic ovarian syndrome, T2DM, multifetal gestations, foetal hydros but in rarer cases in uncomplicated pregnancies. Whilst occasionally seen early in pregnancy, most cases (54%) are diagnosed in the 3rd trimester.

Macroscopic appearances are similar to ovarian hyperstimulation syndrome with both ovaries containing multiple thin-walled theca luteal cysts often mixed with hard residual ovarian stroma. From the outset this can be difficult to distinguish from a bilateral ovarian malignancy and thus at the time of caesarean, surgeons confronted with these findings will often react with unnecessary surgical intervention, which can have profound complications both for fertility and long term health.

Whilst there are no formal studies in the literature, case reports would suggest that ~15-30% of cases are associated with maternal virilisation secondary to hyperandrogenism. Symptoms may include temporal balding, vocal hoarseness, hirsutism and clitoromegaly. Foetal virilisation has been only been reported rarely. Rarely, spontaneous internal haemorrhage into the cysts can present as abdominal pain, emesis and ascites. Laboratory results will often be typified by elevated serum HCG levels, hyperglycaemia, elevated serum androgens as well as an unremarkable tumour marker profile. A study by Haimor-Kochman et al found that MRI did not alter management when compared with pelvic ultrasonography.

Whilst often a benign finding, they have been associated with preeclampsia, HELLP, preterm birth as well as maternal hyperthyroidism. Whilst high levels of HCG are hypothesized to cause placental dysfunction, the exact pathogenesis is not fully understood. It is a self limiting condition and often resolves within 6 months of delivery without any long term maternal or foetal morbidity. Conservative management is therefore ideal.