

Case Report

Large solitary luteinized follicle cyst of pregnancy and puerperium Moawia E Hummeida¹, Ali Abdel Satir², Ali M Ali³, El Rayah M. Mostafa⁴

ABSTRACT

Solitary luteinized follicle cyst is a rare cause of ovarian enlargement during pregnancy and puerperium. Only rare cases of this clinical condition have been reported in the literature.

We report a case of a large solitary luteinized follicle cyst arising in a 32-year-old multipara four days post partum which required postpartum surgical intervention. The perinatal outcome is usually good, with liveborn infants being delivered at or near term. The complications of the disease include ovarian torsion, intra-cystic haemorrhage, and rupture. The condition should be included in the differential diagnostic workup whenever a growing simple cyst is encountered during pregnancy and puerperium.

Key words: luteinized follicle cyst; pregnancy; puerperium.

Ovarian masses are relatively common in pregnancy, with a prevalence of 2.3–5.4% being reported in prospective studies owing to the increase use of ultrasound examination in pregnancy. Functional cysts represent the vast majority of these masses, followed by benign cystic teratomas, serous cystadenomas, paraovarian cysts, mucinous cystadenomas, endometriomas and rarely malignant tumors¹.

The majority of adnexal cysts are small (<5 cm), remain asymptomatic and tend to resolve before birth^{2, 3}, mostly during the first trimester⁴. Simple cysts of > 5 cm in diameter are much less common (representing about 4% of the total). They still tend to resolve spontaneously; however, compared with smaller cysts, they are more likely to develop torsion or haemorrhage, require surgical removal during pregnancy or even be associated with malignancy, especially when

growing^{3,5,6}. A rapidly enlarging ovarian mass in pregnancy poses significant diagnostic problems.

Large luteinized cysts of pregnancy are an uncommon type of cystic mass particular to pregnancy, characterized by the combination of a benign appearance and a tendency to enlarge rapidly, eventually becoming symptomatic and most often necessitating surgery.

PRESENTATION OF CASE:

Our patient was a 32-years-old housewife G5 P4. All deliveries were normal at home, she has two living boys and two girls, and youngest is three years old. She presented to the clinic with a history of vague abdominal pain, gradually increasing discomfort and distension four days following spontaneous full term vaginal delivery at home.

Her medical and family history was non-significant. Her previous obstetric history was normal. She has no regular antenatal care during current pregnancy; she has been seen once by a midwife and has no history of laboratory or ultrasound examination. Pregnancy and delivery were reported as uneventful. She gave birth to a healthy 3.700 g male infant at home four days before admission and she is breast

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feeding. On admission her general condition was good. Her abdomen was grossly distended, soft with mild tenderness and no rebound pain. The remainder of the examination was normal apart from lower limb oedema.

Ultrasound examination revealed a large solitary, unilocular cyst with a thin, smooth wall and homogeneous anechoic content. The cyst has no septae, lacked mural blood flow and was not associated with ascites. It was located in the central abdominal area above and anterior to the postpartum uterus

CT scan report showed a huge pelvi-abdominal cystic structure, arising from the left side of the pelvis and extending upward and lies just below the liver. It displaces the bowel loops aside. It has thin walls, no enhancing septations and not associated calcifications or soft tissue component. The cyst measures 27X16 cm. Uterus is bulky with normal walls with normal urinary bladder. No hydronephrosis or hydroureters. No ascites or enlarged lymph nodes.

Other laboratory tests were normal and CA125 was 5.5 m/u.

Laparotomy was carried out and a right side huge solitary unilocular cyst measuring 26 x 25 x 40 cm was identified (Figure 1). The left ovary was normal; there were no septa, no accompanying ascites, and no other abdominal or pelvic abnormalities were detected. A right Salpingo-oophorectomy was performed. Her postoperative course was uneventful and was discharged home in good condition.

Macroscopic examination shows 6,200 g left ovarian cyst measuring 40 cm in maximum diameter. Cut surface shows unilocular cyst containing turbid straw coloured fluid. Microscopy shows ovarian cyst lined by luteinized granulosa cells (Figure 2). No evidence of neoplasm of adequate sample examined. The features

are those of large solitary luteinized follicle cyst of pregnancy and puerperium.

DISCUSSION:

In spite of a very large cyst (27X16 cm) (6,200 g) which almost fills the entire abdominal cavity, the patient remained asymptomatic during pregnancy, only to be diagnosed in early puerperium by ultrasound due to persistent distension and discomfort.

The pathogenesis of large luteinized cysts of pregnancy is unclear. Its occurrence in pregnancy suggests stimulation with Human chorionic gonadotropin (HCG)⁷, or increased tissue sensitivity to hCG⁷. However, these cysts can also appear up to 3 months postpartum^{8,9}, when HCG has normally become undetectable. Thus HCG may not be the only contributing factor. In such cases, Clement and Scully postulated that the development of these cysts is initially (prenatally) stimulated by HCG, albeit to a size that may remain unnoticed. A rise in the pituitary gonadotropins (follicle-stimulating hormone/luteinizing hormone), if there is no lactation postpartum, may be responsible for their continuing enlargement⁷.

The ultrasound appearance of large luteinized cysts of pregnancy facilitates their differential diagnosis from other, more common, adnexal masses. Follicular and corpus luteum cysts are relatively common, but they rarely exceed 6–8 cm in diameter^{10,11}. Pregnancy luteoma and hyperreactioluteinosis only occur during pregnancy. Both of these masses can cause maternal virilization (25–35%), with luteomas also causing virilization of the female fetus in about one-third of cases^{7,12}. When diagnosed prenatally, they appear as complex ovarian masses. Luteomas are typically multiple and often occur bilaterally. Their appearance is predominantly solid, but they may also have cystic areas and are typically 6–10cm



Figure 1: Left Large solitary luteinized follicle cyst of pregnancy and puerperium.

in size, occasionally reaching 20 cm^{10,13}. Hyperreactioluteinosis is typically bilateral and is characterized by ovarian enlargement caused by numerous luteinized follicular cysts, often with hemorrhagic areas. In contrast to luteoma, hyperreactioluteinosis is seen in conditions with abnormally high levels of HCG (e.g. twins, hydatidiform mole, hydrops and

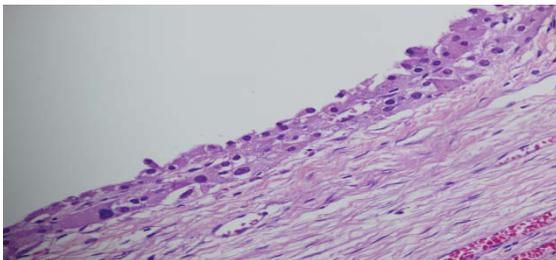
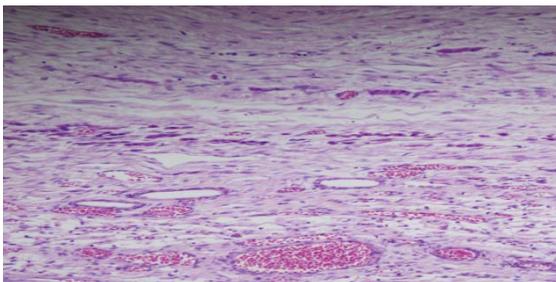
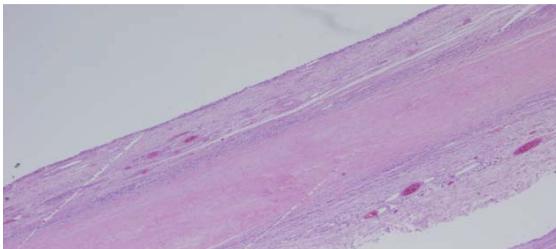


Figure 2: Microscopy shows ovarian cyst lined by luteinized granulosa cells.

choriocarcinoma)^{10,12}.

A diagnostic problem may arise with serous cystadenomas, which are usually unilocular (occasionally bilocular), with a thin, smooth wall, a thin, regular septum (when present) and homogeneous anechoic content¹⁴. The same is true for benign cystic teratomas, which can vary from entirely cystic to echogenic, with different degrees of density and shadowing¹⁵. Such cysts do not usually demonstrate the rapid growth rate reported for large luteinized cysts of pregnancy, and only rarely require removal during pregnancy because of complication¹⁶.

The potential for malignancy is always a concern when considering a rapidly enlarging ovarian mass. Fortunately, less than 1% of ovarian masses in pregnancy are malignant, about one-third of them being germ cell tumors¹⁷, and therefore the positive prognostic value of any ultrasound sign is low. Still, close monitoring would be recommended for large masses, especially as the reliability of ancillary tests (e.g. Doppler indices, CA 125, β -HCG or alpha-fetoprotein) is reduced as a result of the physiological adaptations of pregnancy.

As with any adnexal mass, the management options include either expectant follow-up or surgical removal. Data from series in pregnancy indicate that the risk for torsion in cysts 6–8 cm in diameter is 22%⁶; and, one in nine cysts of more than 5 cm will require surgery and this rate increases to 45% for symptomatic cysts¹⁹. Although published data specifically concerning large luteinized cysts of pregnancy are limited, their tendency to grow means that they are associated with higher risk, and removal was eventually required, either before delivery^{10,19} or during puerperium (as in our case)^{20,21}. Fortunately, it appears that adnexal surgery in pregnancy is relatively safe.

CONCLUSION:

In conclusion, large luteinized follicular cysts of pregnancy are an uncommon type of cystic mass particular to pregnancy, which should be included in the differential diagnostic workup whenever a growing simple cyst is encountered. These cysts are characterized by the combination of a benign appearance and a tendency to enlarge rapidly, eventually becoming symptomatic and most often necessitating surgery.

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