



## Case Report

# A twisted giant corpus luteum cyst in the third trimester of pregnancy in a multigravida: A case report

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## ABSTRACT

Corpus luteum cyst is common in early pregnancy and seldom exceeds 5 cm in diameter; many of which resolve before the end of the first trimester of pregnancy. Giant corpus luteum cyst in third trimester is very rare especially in naturally-conceived pregnancies. We hereby report a rare case of twisted giant corpus luteum cyst in the third trimester in a 33-year-old multigravida. She presented with acute abdominal pain and vomiting at a gestational age of 34 weeks. Ultrasound scan revealed a twisted giant right ovarian cyst. She underwent an emergency cesarean delivery and right oophorectomy. Histology report showed features that were diagnostic of corpus luteum cyst.

**Keywords:** Abdominal pain in pregnancy, Twisted adnexal mass, Persistent corpus luteum

## INTRODUCTION

Small unilateral follicular and corpus luteum cysts are commonly seen in the first trimester during routine ultrasound scan as incidental findings of no clinical significance.<sup>[1-3]</sup> Most of these cysts resolve before the 13<sup>th</sup> week of gestation.<sup>[3]</sup> They rarely persist to the third trimester of gestation and seldom exceed 5 cm in the widest diameter.<sup>[1]</sup> Corpus luteum cysts occur as a result of persistence of the corpus luteum formed following ovulation which secretes progesterone necessary for the maturation of the endometrium in preparation for the implantation of the fertilized ovum.<sup>[4]</sup> In artificially-conceived pregnancies, especially with combined ovarian hyper-stimulation during *in vitro* fertilization procedures; multiple corpus luteum cysts may form in both ovaries and may enlarge as part of ovarian hyper-stimulation syndrome. This is often associated with ascites and other systemic complications.<sup>[5,6]</sup> Unilateral giant corpus luteum cyst persisting in the third trimester following spontaneous conception is a very rare occurrence and has not been previously reported in our tertiary institution.

Torsion of corpus luteum cyst in pregnancy is also extremely rare. The commonly reported cases are those of dermoid cysts and serous cystadenomas that often manifest in the second trimester.<sup>[7]</sup> The solid components and the complex nature of these cysts, coupled with long thin pedicles, make them predisposed to torsion. Torsion of ovarian cyst is a medical emergency due to severe pain and vomiting, which may lead to ischemia of the affected adnexa as well as systemic complications like electrolyte derangement, if not attended to immediately.<sup>[7]</sup> Furthermore, the complete twisting of the cysts results in occlusion of vascular and lymphatic flow which if persists, can lead to necrosis, gangrene, complete loss of reproductive organs, and mortality.<sup>[7,8]</sup>

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The diagnosis is often made from symptoms, clinical examination and confirmed with imaging studies such as ultrasonography with or without Doppler, magnetic resonance imaging, and computed tomography.<sup>[2-5]</sup> Management approach depends on symptoms, gestational age, and among other factors. We present a rare case of twisted giant corpus luteum cyst which occurred in a second ligravida at third trimester with no previous risk factors.

## CASE REPORT

Mrs C.O was a 33-year-old Gravida 2 para 1 (1 alive) lady who had an uneventful antenatal period until the gestational age of 34 weeks. She presented at the Calabar Hospital with acute abdominal pain and vomiting of 1 day duration. She had no fever, bone pain, dizziness, urinary symptoms, or fainting spells. She still felt regular fetal movements. There was no vaginal bleeding or abnormal vaginal discharge. The pain was described as sharp and radiated to the lower back and upper right thigh. There was no diarrhea or constipation. There was no antecedent history of easy fullness, dyspepsia, epigastric pain, or abdominal trauma.

She had a regular 28-day menstrual cycle and the average flow duration was 4 days. She conceived spontaneously. The early ultrasound scan (done on February 4, 2020) showed a singleton pregnancy with sonographic gestation age of 9 weeks (from crown-rump length) equivalent to her gestational age from last menstrual period. She was not on any hormonal medication before conception. Her first child was 2 years and 4-months-old, male, and was delivered by cesarean section due to abnormal lie at term.

On general physical examination: She was a young lady, conscious and in painful distress. She was not pale and had no pitting pedal edema. Her pulse rate was 98 beats/min and the pulse volume was adequate and regular. Her systolic blood pressure was 129 mmHg while diastolic was 80 mmHg. Her respiratory and pulse rates were within normal limits. The abdominal examination revealed a uniformly enlarged abdomen with generalized tenderness and guarding. The uterine size corresponded with her gestational age of 34 weeks. The fetal heart tones were present. Vaginal examination revealed a closed cervix. Urgent abdominal ultrasound scan done revealed a live male fetus in longitudinal lie and cephalic presentation. The sonographic estimated fetal weight was 2.4 kg. The placenta was fundally-sited with no evidence of retro placental blood clots. There was a right adnexal mass, circular, thin-walled, and containing clear fluid. Its widest diameter was 32 cm [Figure 1]. The capsule was smooth. There was no mass in the left adnexa and no ascites. A diagnosis of twisted giant ovarian cyst in pregnancy was made.

Her full blood count, electrolyte, urea and creatinine, and urine analysis were all within normal range. She was

immediately prepared for emergency cesarean section and right oophorectomy due to acute ovarian torsion in advanced pregnancy.

The peritoneal cavity was entered using the old low transverse supra-pubic incision. The indication for the previous cesarean section was abnormal fetal lie at term. Intraoperatively, the baby was first delivered. The weight of the baby was 2.3 kg and the APGAR score was 8/10 and 9/10 in 1 and 5 min, respectively. After the repair of the uterine wound, the uterus was exteriorized and the twisted right adnexa delivered as shown in Figure 2. The large cyst was manipulated and delivered *en bloc* through the cesarean incision as shown in Figure 2, the right oophorectomy was then performed. The delivered cyst was intact as shown in Figure 2. The abdominal incision was closed in a usual way after thorough inspection and palpation of the abdominal and pelvic structures including the contralateral adnexa.

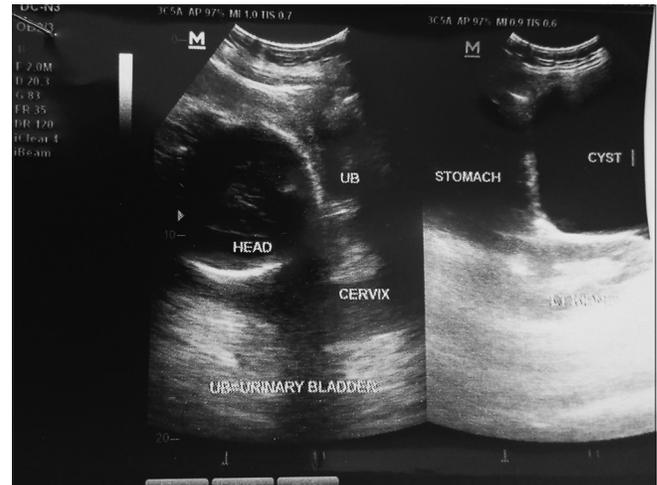


Figure 1: Imaging of the giant cyst extending into the epigastrium.



Figure 2: The giant cyst twisted at its pedicle.

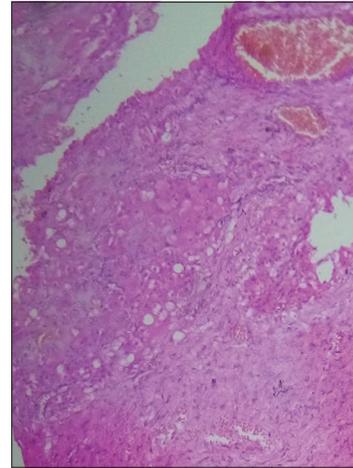
Her post-operative period was uneventful and the baby was commenced on exclusive breastfeeding. The histopathology result was consistent with the diagnosis of corpus luteum cyst. She was discharged from the hospital on the 3<sup>rd</sup> post-operative day in good condition and given a follow-up appointment.

## DISCUSSION

The ovary is a common site for cyst formation in women during their reproductive years. In pregnancy, the incidence of torsion of ovarian cyst is reported at 5 in 10,000 pregnancies.<sup>[3,8,9]</sup> Histologically, the commonly reported twisted ovarian cysts during pregnancy include serous cystadenomas, teratomas, and endometriomas. The predisposing factors for torsion in these tumors include: The size of cyst, long pedicle, mobility, and the internal contents of the cysts.<sup>[10,11]</sup> They often present between 14 and 28 weeks of gestation.<sup>[7]</sup> Twisted giant corpus luteum in the third trimester is a rare incidence. Its attachment to the ovary is broad and the torsion usually likely to involve the whole unilateral adnexa. Cystectomy is difficult implying that oophorectomy is often indicated.

The corpus luteum in pregnancy is supported and maintained by human chorionic gonadotropin (HCG). Enlargement of corpus luteum cyst has been linked to ovarian hyperstimulation syndrome (OHSS) during assisted reproduction procedures.<sup>[12]</sup> OHSS can be caused by high levels or increased sensitivity to follicle stimulating hormone (FSH) or HCG.<sup>[13,14]</sup> In the literature, OHSS has been reported due to FSH secreting gonadotrope cell adenoma and the symptoms resolved following transsphenoidal tumor resection.<sup>[15]</sup> Several FSH receptors mutations have been identified as rare causes of ovarian hyper-stimulation.<sup>[15]</sup> The possible etiological factor in this patient is unclear. She had no clinical evidence of pituitary adenoma and FSH receptor hypersensitivity or mutation could not be completely excluded.

Twisted ovarian cyst in pregnancy is associated with significant maternal and perinatal morbidity and mortality if timely intervention is not instituted.<sup>[6-8]</sup> Accurate clinical assessment for proper diagnosis is an essential component of management. Diagnoses which are often inferred from the acute symptoms are usually confirmed with imaging studies such as ultrasonography, computed tomography, and magnetic resonance imaging. Their sensitivity and specificity are comparable. Intraoperative Frozen section may be necessary to exclude malignancy and determine the extent of surgical resection. It is important to subject every specimen to histological analysis irrespective of the clinical picture. In this patient, the histopathological analysis revealed cells with abundant clear cytoplasm with prominent round to oval nuclei with underlying fibrocollagenous theca layer [Figure 3]. This was consistent with corpus luteum cyst.



**Figure 3:** Section of a cyst wall lined by luteinized granulosa cells (with abundant clear cytoplasm and a prominent round to oval nuclei).

The surgical approach may be laparoscopy or laparotomy depending on patients' clinical condition, the size of the cyst, presence of any co-morbidity, and the gestational age at presentation. Laparoscopy has the benefits of less blood loss, cosmesis, shorter duration of hospitalization, and better post-operative pain management when compared to laparotomy.<sup>[14]</sup> However, the risk of resorting to open laparotomy is significant especially when malignancy cannot be completely rule out. The incidence of malignancy with persistent adnexal tumors is reported between 2% and 6%. For large adnexal masses in the third trimester, laparotomy is the preferred approach as in the index case.

Caesarean oophorectomy or cystectomy is a rare procedure in the management of adnexal masses in pregnancy. In this patient, the baby was delivered by cesarean section before the twisted mass which was posterior to the uterus was accessed, delivered *en bloc* and oophorectomy performed. In the literature one, 14 year case series showed that majority of the twisted adnexal masses occurred in the second trimester with mean gestational age of 14 weeks at presentation.<sup>[7,11]</sup> In most of these cases reported, the pregnancy progressed to full-term delivery of live infant with an average gestational age of  $37.78 \pm 3.42$  and mean birth weight was  $2.97 \text{ kg} \pm 0.65$ .<sup>[7]</sup> The method of treatment adopted for this patient was justifiable due to several factors: First, it would have been technically difficult to access the twisted adnexa with the baby in the uterus; second, the baby was delivered at a gestational age of 34 weeks and sonographic estimated fetal weight of 2.4 kg; third the woman had one previous cesarean section which is a relative indication for a repeat section; and finally large ovarian cyst can cause difficult labor if the fetal part is obstructed in the pelvic cavity. Consecutive management was contraindicated in this case. The acute pain and the risk of severe maternal complications as a result of ischemia and gangrene from the twisted adnexal mass made conservative management unsuitable.

## CONCLUSION

Twisted corpus luteum cyst in pregnancy is a rare obstetric emergency requiring prompt intervention to prevent maternal and perinatal mortality and morbidity. When diagnosed in the third trimester, emergency cesarean cystectomy is a treatment of choice.

## ACKNOWLEDGMENTS

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## ETHICAL APPROVAL

Ethical approval was obtained from the management of Calabar Hospital.

## AUTHORS CONTRIBUTIONS

Author 1 was involved in the management of the patient, Author 2 supervised the histologic analysis while Author 3 assisted in the final write-up.

## Availability of data

The case report is available to be shared with the permission of the authors.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

## Conflicts of interest

There are no conflicts of interest.

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