



Giant ovarian mucinous cystadenoma complicating term pregnancy: a rare case report

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Introduction: Adnexal masses in pregnancy are identified at a rate of 2 to 20 in 1000, which mostly are benign simple cysts. Although the majority of adnexal masses will resolve spontaneously by the beginning of the second trimester, some cases are persistent forms which can result in complications for both mother and fetus.

Presentation of case: A 31-year-old pregnant woman was diagnosed with a left ovarian mucinous cystadenoma. The ovarian cyst was first detected in the 8th week of pregnancy, measured 5 × 5 cm and reached 40 × 30 cm at 38 weeks of gestation. The patient underwent an elective cesarean delivery along with a left salpingo-oophorectomy at term. An alive female baby of 3000 g was delivered with good APGAR scores at the first and fifth minutes. Histologic examination confirmed the diagnosis of a benign mucinous cystadenoma.

Discussion: The option of surgical intervention versus conservative management should be tailored to the patient's physical symptoms, characteristics of the tumor, and gestational age. An elective cesarean section along with salpingo-oophorectomy can be adopted for a giant ovarian cyst complicating term pregnancy. There is a risk of recurrence after a removal of ovarian mucinous cystadenoma.

Conclusion: Ovarian cysts coexisting with pregnancy should be followed up properly by routine ultrasound and regular antenatal care. The decision to postpone surgical management of a complex mass until the time of delivery must balance the risks and benefits for mother and fetus. After surgery, the patient needs to be provided an appropriate follow-up care to manage postoperative recurrence.

Keywords: adnexal mass, case report, ovarian mucinous cystadenoma, pregnancy

Introduction

Adnexal masses are diagnosed in 0.2 to 2% of pregnancies^[1], which mostly are benign simple cysts with a diameter of less than 5 cm^[2,3]. Ovarian cysts coexisting with pregnancy are usually asymptomatic and incidentally detected in the first-trimester ultrasound examination. However, some giant ovarian cysts may present with abnormal symptoms such as urinary retention, abdominal or back pain, bloating, vomiting, and constipation^[4].

Most adnexal masses will resolve spontaneously by the beginning of the second trimester, which aligns with the natural progression of functional cysts^[5]. However, some cases are persistent forms which can result in complications for both mother

HIGHLIGHTS

- Giant ovarian mucinous cystadenomas are rare in pregnancy, but can result in complications for both mother and fetus.
- An elective cesarean section along with salpingo-oophorectomy can be adopted for a giant ovarian cyst complicating term pregnancy.
- Ovarian cysts coexisting with pregnancy should be followed up properly by routine ultrasound and regular antenatal care.
- After surgery, the patient needs to be provided an appropriate follow-up care to manage postoperative recurrence.

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Sponsorships or competing interests that may be relevant to content are disclosed at the end of this articles.

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Annals of Medicine & Surgery (2024) 86:6819–6823

Received 6 August 2024; Accepted 16 September 2024

Published online 25 September 2024

<https://dx.doi.org/10.1097/MS9.0000000000002606>

and fetus, including adnexal torsion, cystic rupture, hemorrhage, infection, obstructed labor, fetal malpresentation, abortion, or preterm labor^[5,6]. The majority of benign adnexal masses during pregnancy are physiological cysts (follicular, corpus luteum, and theca lutein cysts)^[7]. Other benign tumors include mature cystic teratoma, serous, and mucinous cystadenoma. The mucinous cystadenomas are epithelial ovarian tumors, which tend to be unilateral and multilocular with smooth surface and contain mucinous fluid.

So far, there is no a certain consensus regarding the management of adnexal masses during pregnancy^[8]. While some obstetricians prefer the conservative treatment because these lesions can resolve with progressing gestation, others decide to perform elective surgery in second trimester to prevent complications. The

option of expectation management versus surgical intervention should be tailored to the patient's physical symptoms, characteristics of tumor, and gestational age.

Although ovarian mucinous cystadenoma can grow rapidly to an enormous size and result in adverse fetomaternal outcomes during pregnancy, there are limited published data investigating the progression of ovarian mucinous cystadenoma in pregnant women. In this report, we present a case of giant left ovarian mucinous cystadenoma that was diagnosed in the third-trimester pregnancy and surgically removed during a cesarean section. This case report has been reported in line with Surgical CAse Report (SCARE) guidelines^[9].

Presentation of case

A 31-year-old pregnant woman (gravida 1, para 1) was admitted to our hospital at 38 weeks of gestation in February 2024. She had regular menstrual cycles, and the determination of gestational age was based on her last menstrual period. She underwent a cesarean delivery in 2012 and a laparoscopic removal of giant ovarian mucinous cystadenoma measuring 20 × 30 cm in 2023.

Three months after ovarian cystectomy, the patient got pregnant. At 8 gestational weeks, a singleton live fetus and a simple cystic 5 × 5 cm mass in the left abdominal area were found by ultrasound. The patient was monitored at a local antenatal clinic. Upon arrival at our hospital, she was at 30 weeks of gestation with a septated cystic mass measuring 23 × 15 cm in the left adnexa with a normal level of tumor markers. The patient and her relatives were informed about her current situation and potential complications. After discussion, they decided to choose conservative management until full term and the patient was kept on regular follow-up at our hospital. No complication was reported during the third-trimester of pregnancy. The patient was hospitalized for an elective cesarean section at 38 weeks of gestation.

After hospitalization, a general examination revealed normal vital signs except for shortness of breath when lying down. The abdomen was grossly distended, with a huge mass arising from the pelvis up to the xiphisternum. The presentation of the fetus and uterine contraction could not be appreciated. There was no bleeding or discharge, and vaginal examination suggested a closed cervix. Laboratory investigations, including complete blood count, serum biochemistry, coagulation tests, and tumor markers of cancer antigen-125 (CA-125) and human epididymis protein 4 (HE4) were within normal limits. Her alpha-fetoproteins (AFP) level was 143 ng/ml, which was higher than normal range (0–10 ng/ml). Ultrasonography showed a large multiseptated, smooth surfaced cystic 40 × 30 cm mass at the left side of the uterus with no solid component or papillary projections. The Ovarian-Adnexal Reporting & Data System (O-RADS) score evaluated by the imaging specialist was O-RADS 3, corresponding to a very low malignant risk. The patient underwent an abdominal MRI scan to fully evaluate the tumor before surgery. MRI revealed a cystic structure of 35 × 30 × 13 cm in diameter with well-delineated, thin walled, and homogeneous signal in the left abdominal area, exerting pressure on adjacent structures (Fig. 1).

The patient underwent an elective cesarean delivery due to previous cesarean section and the presence of a large abdominal mass. Laparotomy was performed through a midline skin incision and a transverse lower uterine segment incision. An alive female

baby of 3000 g was delivered with good APGAR scores at the first and fifth minutes. Intraoperative findings were extraordinary large multiloculated left ovarian cyst measuring 40 × 30 cm, weighing 6 kg with 5 l of straw-colored mucinous fluid content, occupying the entire pelvis and abdomen (Fig. 2). The external surface of the cyst was smooth and pinkish, however, inner surface contained multiple septa, but no solid component or hemorrhagic areas were seen. There was no adhesion to the surrounding tissues. The right ovary and other pelvic organs were normal in appearance. A left salpingo-oophorectomy was done along with right-sided tubal ligation. The patient made an uneventful recovery and was discharged from the hospital on postoperative day 7. Histologic examination of the surgical specimens revealed a benign mucinous cystadenoma, as shown in Figure 3.

Discussion

Ultrasound is the mainstay of diagnosis of adnexal masses, with a sensitivity of 96.8% and a specificity of 77%^[10]. The detection of adnexal masses in pregnancy has become commonplace due to the widespread use of prenatal ultrasound. However, accurately differentiating between benign and malignant adnexal masses through ultrasound remains challenging. One approach to determine the likelihood of malignancy in an adnexal mass on ultrasound is described by the International Ovarian Tumor Analysis (IOTA) study group. This is based on a set of five ultrasound features indicative of a benign tumor (B-features) and five ultrasound features indicative of a malignant tumor (M-features). There are no sonographic features that differentiate an ovarian borderline tumor from a benign or invasive ovarian neoplasm. When in doubt, further evaluation by MRI scan can help to distinguish benign from malignant tumors, with an overall accuracy of 88–93%^[11]. Ultrasonography, in this case, revealed a large benign adnexal mass at the left side of the uterus. We performed an MRI scan to identify exactly the origin, location, and size of the tumor. The advantage of MRI over ultrasound is that it can visualize the more posterior and lateral area of the pelvis, which may be obscured by fetal bones. In this case, an IMR scan demonstrated a giant benign tumor probably originating from the left ovary.

Tumor markers such as CA-125, HE4, and AFP show little clinical significance during pregnancy because their serum levels are normally elevated in pregnant women. In our study, apart from elevated AFP levels, the rest of the markers, including CA-125 and HE4, were within the normal range. It is well-known that maternal serum AFP level begins to increase from 14 weeks of gestation and reach a peak at 32 weeks. First, AFP is produced by the yolk sac, then by the fetal liver and gastrointestinal tract^[12]. During pregnancy, AFP is routinely used for fetal surveillance, particularly neural tube defects and trisomy 21, rather than tumor detection^[13].

The therapeutic approach to adnexal masses changes according to tumor size, gestational age, and clinical presentation. The main consideration in choosing surgical intervention versus conservative management must center on the risks to the mother and fetus. Expectant management of asymptomatic corpus luteal cysts, endometriomas, and mature teratomas (dermoid) during pregnancy is appropriate if the diagnosis is reasonably certain based on the sonographic characteristics. Surgical intervention is

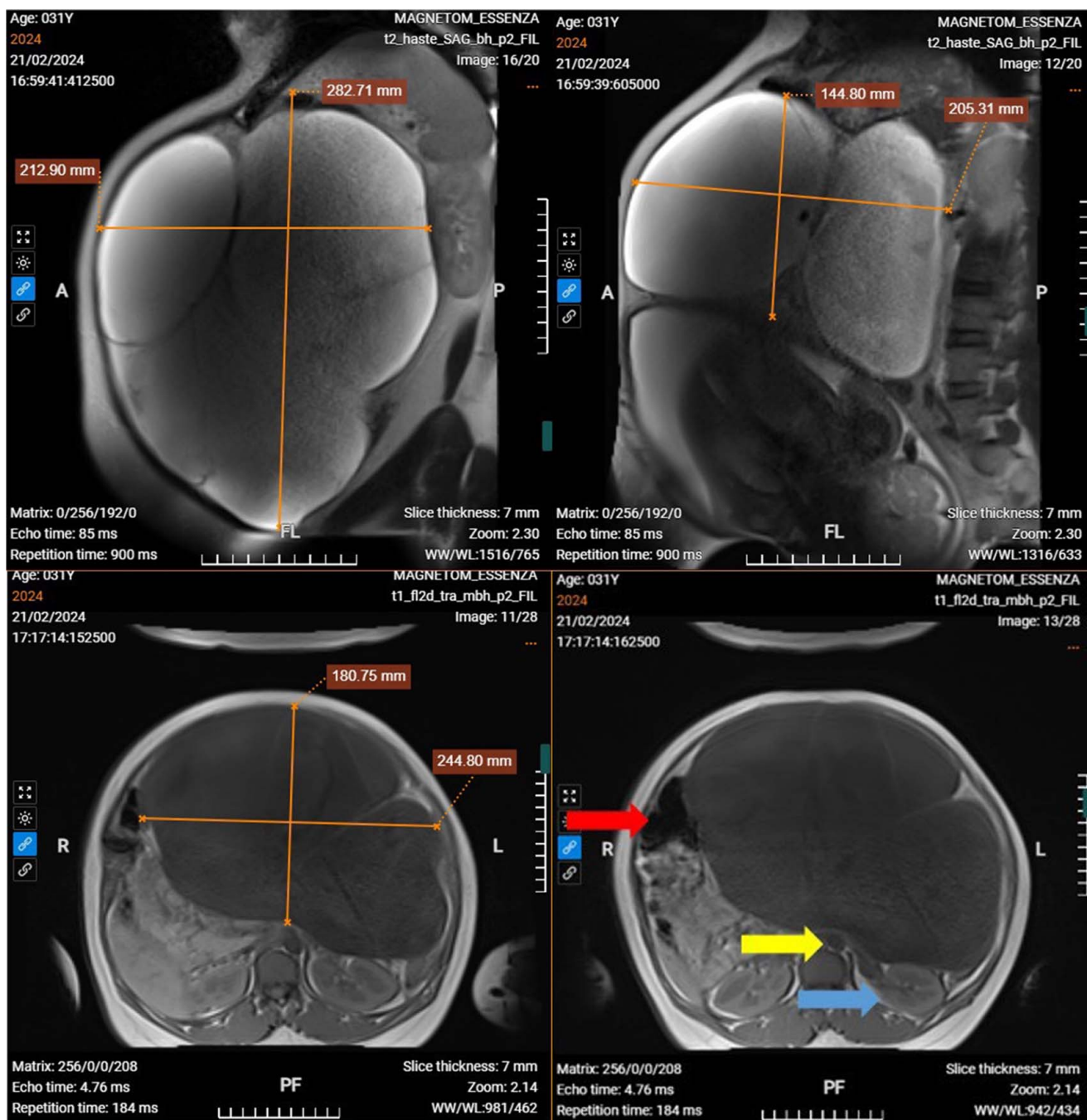


Figure 1. Abdominal MRI showed a giant ovarian tumor at 38 weeks of pregnancy that pressed on adjacent structures (red arrow: colon corner, green arrow: left kidney, yellow arrow: aorta).

recommended for ovarian masses that have at least one of the following features: (1) a strong suspicion of malignancy and/or large size ($> 8-10$ cm), (2) symptomatic complaints, or (3) an increased risk of torsion/rupture/obstruction of labor^[10].

There is a consensus that the best time for surgery is second trimester^[14]. Performing surgery in the first-trimester may increase the risk of spontaneous abortion and luteal dysfunction, whereas intervention in the third-trimester can cause some major complications such as torsion, rupture, hemorrhage, intrauterine growth restriction, or preterm birth. In this case, the patient had a

history of removal of giant ovarian mucinous cystadenoma, and her recurrent ovarian mucinous cyst was first detected at 8 weeks of gestation. However, the patient was not referred to our hospital until 30 weeks of gestation. If the patient's medical history was known and highlighted during the initial 8-week ultrasound, a referral to our hospital may have been earlier during the second trimester, and we can offer a more appropriate treatment for the patient. This is a clinical experience we draw from our case study and want to share with young practitioners. In addition, the late antenatal care was partly attributable to poor economic support,

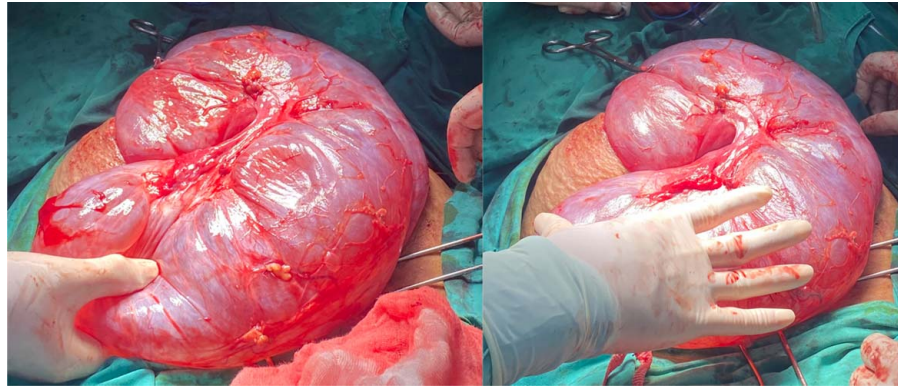


Figure 2. Macroscopic appearance of the tumor during surgery.

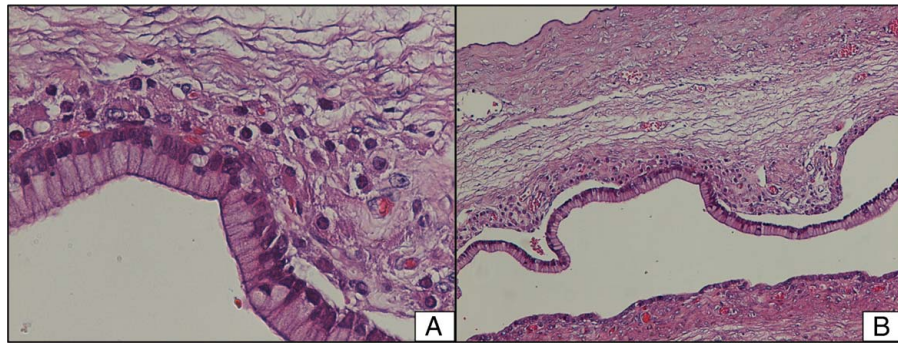


Figure 3. Microscopic appearance of the tumor: the tumor is lined by a single row of mucinous epithelium. (A) Observation at 40x magnification. (B) Observation at 10x magnification.

long commuting distances, multi-child family, unaware of pregnancy-related danger signs, and inappropriate perception of antenatal care starting time^[15,16]. Although this patient and her relatives were informed about the potential complications of delaying treatment, they still decided to postpone the operation until the end of week 37. There are several case reports in the literature that considered expectant management as the best approach for a huge ovarian cyst presenting in the third-trimester and removed the cyst at the time of elective caesarian section^[17–19]. Meanwhile, Qublan *et al.*^[17] and Yenicesu *et al.*^[19] presented two cases of huge ovarian mucinous cystadenomas complicated with intrauterine growth restriction at term, our case showed a normal birth weight of 3000 g at 38 weeks of gestation.

The choice of laparotomy versus laparoscopy is dependent on the risks of malignancy, the urgency of the procedure, and the skills of the surgeon^[10]. Laparoscopy has been consistently shown to be associated with shorter hospital length of stay and less postoperative pain and can be associated with less manipulation of pelvic organs and better surgical exposure^[20]. However, laparoscopy is not always feasible at later gestational ages owing to the impact of uterine size on intra-abdominal visualization and impeded access to pelvic structures. In this case, the patient was taken for an elective cesarean section along with left salpingo-oophorectomy at 38 weeks of gestation. We used midline incision because this incision provided the best visualization and intra-abdominal access with little blood loss^[21]. The left sal-

pingo-oophorectomy was performed as there was no normal ovarian tissue left, and the fallopian tube was damaged.

Our patient experienced a recurrence of a left ovarian mucinous cyst during pregnancy. She underwent a removal of a giant left ovarian mucinous cystadenoma by laparoscopic cystectomy 1 year ago. Although the cystectomy was performed without intraoperative rupture and spillage, the ovarian mucinous cystadenoma still recurred on the left adnexa. We speculate that the recurrence may be due to the remanence of the mucinous tumor cells on the left adnexa from the first operation. Another mechanism for the recurrence and rapid growth of ovarian mucinous cystadenoma during pregnancy could be due to the fact that this type of tumor is hormonally responsive^[17,19]. There are limited data concerning recurrent mucinous cystadenoma following laparoscopic cystectomy during pregnancy. Sanaullah *et al.* presented a case of recurrent mucinous cystadenoma following oophorectomy in the second trimester of two consecutive pregnancies. It was supposed that the newer cyst developed from the presence of residual ovary or ovarian tissue or cyst wall in the stapled ovarian stump^[22]. Further research is needed to better understand the impact of pregnancy on the recurrence of ovarian mucinous cystadenoma. Our case study highlighted the necessity of follow-up for women after cystectomy to reduce the risk of recurrence. Transvaginal ultrasound recommended every 3–6 months seems currently to be the most effective diagnostic tool for the follow-up of young patients treated with cystectomy for benign mucinous cystadenomas^[23].

Conclusion

Ovarian cysts coexisting with pregnancy should be followed up properly by routine ultrasound and regular antenatal care. Ultrasound remains the most important tool in the workup of adnexal masses because of its safety, availability, and accuracy. Antenatal care is necessary to early identify adnexal masses and provide timely interventions during pregnancy. The decision to postpone surgical management of a complex mass until the time of delivery must balance the risks and benefits, weighing the risks of malignancy vs the potential for unnecessary surgical risk for the mother and fetus. After surgery, the patient needs to be provided an appropriate follow-up care to manage postoperative recurrence.

Ethical approval

Patient anonymity is maintained throughout this manuscript, and consent was obtained for publication from the patient.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Source of funding

Not applicable.

Author contribution

N.M.T., N.T.H.A., and H.T.Q.: conceptualization and methodology; N.T.H.A., N.M.T., H.T.Q.: writing – original draft and visualization; N.M.T. and N.T.H.A.: supervision, writing – review and editing. The final manuscript was approved by all authors.

Conflicts of interest disclosure

The authors declare no conflicts of interest.

Research registration unique identifying number (UIN)

Not applicable.

Guarantor

Nguyen Manh Thang.

Data availability statement

Not applicable.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Acknowledgement

Not applicable.

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