EIGHTEEN-YEAR-OLD NULLIPAROUS WOMAN WITH MASSIVE MUCINOUS CYSTADENOMA IN PREGNANCY: CASE REPORT

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Abstract

Background: Ovarian mucinous cystadenoma is rare in adolescents. We report the diagnosis and management of a massive ovarian mucinous cystadenoma in a pregnant adolescent.

Case Presentation: An 18-year-old G1PO was referred by a midwife to the Komfo Anokye Teaching Hospital on account of excessive abdominal distension at 25 weeks' gestation and an ultrasound scan finding of a large non-septate cystic adnexal mass. She, however, reported to the hospital 6 weeks later with a complaint of worsening abdominal distension and discomfort. A transabdominal ultrasound scan revealed single intrauterine gestation and a large non-septate cystic adnexal lesion measuring more than 19 cm in its widest plane. She had a conservative in-patients management followed by emergency caesarean delivery, exploratory laparotomy and unilateral salpingo-ophorectomy at 33 weeks 1 day. The outcome of the delivery was a 1.8 kg female infant with Apgar score of 4 and 6 at 1 and 5 minutes respectively. A 6.8 kg cystic ovarian mass measuring about 40 cm at its widest diameter was removed.

Conclusion: Although pregnancy-associated adnexal masses are not uncommon, significant socio-economic challenges may render the diagnosis and treatment of extremely large pelvic masses in pregnant adolescents difficult. The decision to optimize maternal and fetal outcomes is essential in managing rare conditions in marginalized obstetric populations.

Key Words: *Adolescent Pregnancy, Adnexal tumours, Mucinous cystadenoma, Antenatal care & Conservative surgery*

Introduction

Adnaexal masses can be found in both pregnant and non-pregnant women of any age, with prevalence of 0.19-8.8%.[1] Only 3-6% of all these masses are malignant, and the detection of asymptomatic and clinically in-apparent adnexal lesions are on the increase. [2] This is largely due to increased access, coverage of antenatal care and the availability of prenatal ultrasonography. Most of these lesions are diagnosed in the first trimester, and the incidence decreases with increasing gestation.[3] The benefits of this shift in the practice are not fully realized in the rural and the peri-urban areas of the West African sub-region due to delayed entry into antenatal clinics, logistics and human resource factors in the maternal health system.[4,5] They are consequently diagnosed late, at enormous sizes in late trimesters. Further, antenatal care in the adolescent age is characterized by poor attendance and inadequate care.[6] The underlying factors range from poor economic support, long commuting distances, unfriendly antenatal care programmes to the lack of insight into the usefulness of the care. The set-up makes it even more challenging to diagnose and treat these masses among adolescents in the low- and middle-income countries (LMIC).[7]

Functional cysts (follicular, corpus luteum and theca lutein cysts) account for the majority of benign adnaexal masses in pregnancy.[8] Other benign tumours include mature cystic teratoma, serous and mucinous cystadenoma. The ovarian mucinous cystadenoma is often a multilocular cyst with smooth outer and inner surfaces. Benign ovarian mucinous cystadenoma are common between the third and the fifth decades, but rare at the extremes of age, including the adolescent age.[9] Although they can grow to an enormous size, literature is scanty on rapidly growing ovarian mucinous cystadenoma in a pregnant adolescent.

There is no agreement among authors regarding the definitive management of adnexal masses in pregnancy.[10] Assessment using clinical, biochemical and ultrasonographic indices are helpful in choosing appropriate management recommendations. While some obstetricians prefer the conservative non-surgical approach because majority of these lesions resolve with increasing gestation, others prefer elective surgical treatment in the second trimester with

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concerns of ovarian malignancy, torsion, rupture, entrapment or obstruction of labour.[8] The considerations for the definitive management of ovarian tumours during pregnancy are strongly influenced by clinical, biochemical and imaging findings suggestive of ovarian malignancy or the decision to maximize maternal and fetal outcomes. Conservative surgery (ovarian cystectomy or salpingo-oophorectomy) often suffices as a treatment for most of the benign ovarian tumour.

We, therefore, present a case report of a massive ovarian mucinous cystadenoma in a pregnant adolescent.

Case Report

An 18-year-old nulliparous woman was referred from a peripheral healthcare facility to Komfo Anokye Teaching Hospital (KATH), on account of a suspected ovarian tumour in pregnancy.

The patient made an unusual early visit to the antenatal clinic (ANC) in the first trimester on account of signs and symptoms of anaemia. A full blood count and peripheral blood film assessment reported a haemoglobin level of 6.1 g/dL and a microcystic normochromic picture, respectively. She was treated with haematinics and dietary modifications. A dating ultrasound scan reported a single intrauterine gestation at 8 weeks 3days with no comment on the adnaexa. At a scheduled antenatal visit, the midwife noticed unusual distension of the abdomen. Follow up ultrasound scan reported a large non-septate cystic adnexal mass and appropriately grown fetus at 25 weeks + 6 days. Therefore, she was referred to KATH, but she defaulted and reported later to KATH at 31 weeks + 3 days gestation with worsening abdominal distension and discomfort. Apart from a low haemoglobin level, all the other screening investigations at the booking visit were normal.

She had had regular menstrual cycles prior to pregnancy, and she had never used any form of modern contraception. She had no significant previous medical or surgical history. The family history was negative for ovarian, colon, cervical, breast and endometrial carcinoma. She lived in a rural area, was unemployed and married to a labourer. She had an active health insurance card.

General physical examination revealed normal vital signs except for tachypnoea (respiratory rate 24 cycles/minute). Her weight and height were 64 kg and 162 cm respectively. Her breasts were bilaterally symmetric, with normal looking nipples with prominent areolar. The abdomen was grossly distended, with tribal marks running radially from the umbilicus (Figure 1). There was an obvious bulge in the suprapubic region, and laterally to the flanks bilaterally. The abdomen was tense but non-tender in all regions. The fetal heart rate was 158 beats per minute using sonicaid. The pelvic examination revealed a normal looking vulva, vagina and cervix. There was fullness in the cul-de-sac. It was difficult assessing the pelvic side walls.

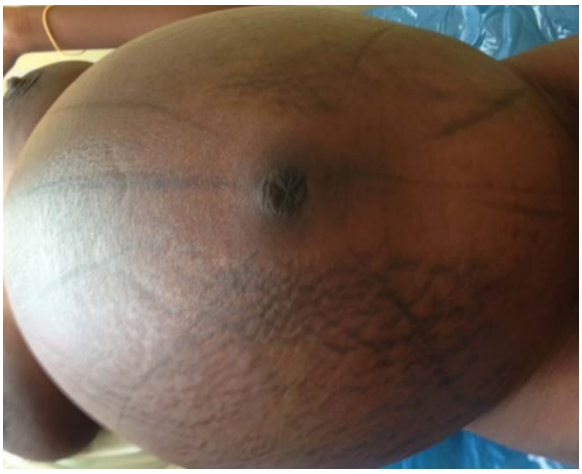

Figure 1: A massively distended abdomen with a bulge in the suprapubic region.

An immediate transabdominal ultrasound scan reported a non-septate cyst with internal echoes (Figure 2). The cystic lesion appeared to originate from the right adnaexum. It measured more than 19 cm in its widest plane. The left ovary could not be visualized. There was minimal free intraperitoneal fluid.

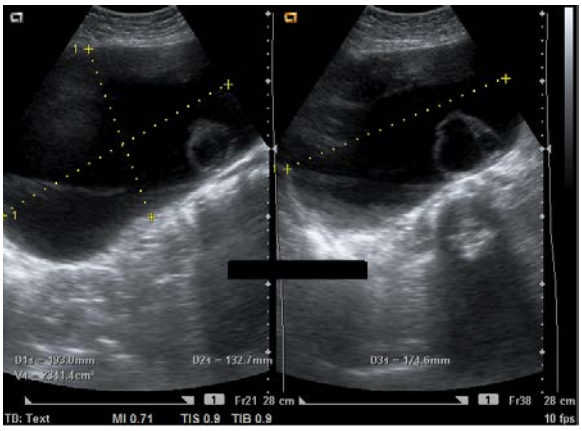

Figure 2: An ultrasound scan of a non-septate ovarian cyst with internal echoes.

Laboratory investigations including full blood count, serum biochemistry, cancer antigen-125 (Ca-125), Latic acid dehydrogenase (LDH) and alpha-fetoproteins (AFP) reported the following findings: normochromic microcytic anaemia with haemoglobin level of 9.3 g/dL, platelet of 240 x 10^3 / microlitre, LDH 349 U/L (225-450), CA-125 29U/ml (0-39), alpha-fetoprotein 123.3U/ml (0-10), and normal liver and renal function tests.

The case was discussed by a multidisciplinary team (MDT) consisting of physicians, nurses and midwives from feto-maternal, gynaecologic oncology, anaesthesia, paediatric and radiation oncology units.

The findings on evaluation were more suggestive of a benign lesion. The decision was to deliver at term by c-section, followed by exploratory laparotomy and treatment of the ovarian tumour. The probability of preterm delivery was also anticipated in this case. She was given dexamethasone to accelerate fetal lung maturation and put on folic acid and iron supplementation. A blood sample was also taken and cross-matched against four units of her blood type and saved at the blood bank. The findings and the management plan were explained to the patient, and informed consent sort for the in-patient care on the antenatal ward. A biweekly non-stress test (NST) was done as part of antepartum fetal monitoring during the in-patient care.

On the tenth day of admission, at 32 weeks + 6 days, she started experiencing mild to moderate lower abdominal pain. The pain was constant with no aggravating factors, and the pain was relieved after taking tramadol injection. This was not associated with vaginal bleeding or show. She could perceive fetal movements. It was not possible to assess contraction by palpation. The findings on vaginal examination were as follows: the cervix was closed, 2cm long, firm and posterior. An urgent ultrasound scan reported an adequate for gestation fetus with normal biophysical profile and umbilical artery velocimetry findings. The uterus was displaced to the left flank, and the nature of the lesion was confirmed as non-septate cystic with internal echoes. The lesion extended to the epigastrium and both hypochondria. The liver had a normal echo-pattern, and there was moderate bilateral hydronephrosis. The NST tracings were normal.

The lower abdominal pain became severe on the twelfth day of admission (33 weeks + 1-day gestation), and this time was associated with vomiting. On general physical examination, she looked agitated. She was moderately pale, not jaundiced and had a temperature of 36.7C. Her blood pressure was110/70mm/Hg and she had a pulse rate of 92 bpm, regular with good volume. Her chest was clinically clear. Her abdomen was grossly distended, very tender and tense with guarding in the umbilical region. Uterine contractions could not be assessed. The fetal heart rate was 148 bpm and was regular. Vaginal examination revealed a central, 2cm long cervix with closed external os.

A diagnosis of suspected ovarian cyst rupture was made. She was immediately prepared for emergency laparotomy and caesarean section. The neonatology team was informed to receive the baby. Under general anaesthesia, an abdominal midline incision was made from the suprapubic region to the epigastrium (Figure 3). The lower uterine segment was poorly formed; hence J-uterine incision was made in the lower segment to deliver the baby, placenta, and membranes. The uterus was repaired in layers (Figure 4).

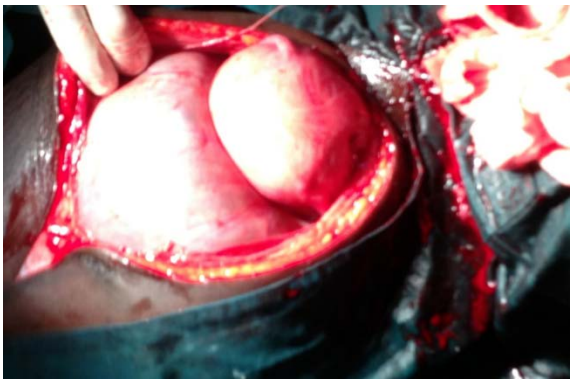

Figure 3: The large cyst and the gravid uterus during the surgery.

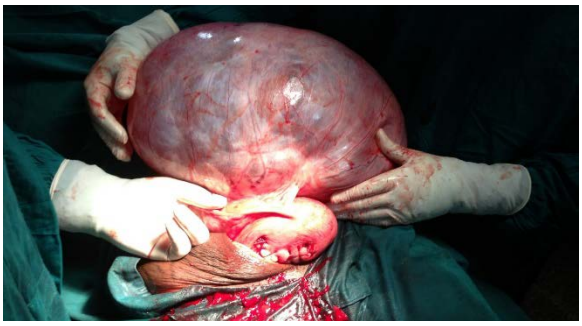

Figure 4: Intact ovarian cyst and the gravid uterus after the caesarean section.

The findings of the surgery were 1.80 kg female infant with Apgar score of 4 and 6 at 1 and 5 minutes respectively; and a 6.80 kg right cystic ovarian mass with an intact capsule. The surface of the ovary was smooth, with no external growths or adhesions (Figure 5). The ipsilateral fallopian tube was elongated and firmly attached to the capsule of the ovarian cyst.
Other intraoperative findings were:

- Grossly normal appearing left ovary and tube.
- Extensively stretched right infundibulopelvic and broad ligament.
- There was no ascites.
- The liver, bowels and the omentum felt normal and had no suspicious lesions.
- No tumor nodules were seen in the parietal and visceral peritoneum, including the Pouch of Douglas (POD), paracolic, subphrenic and subhepatic spaces.
- Pelvic and paraaortic lymph nodes were not enlarged.

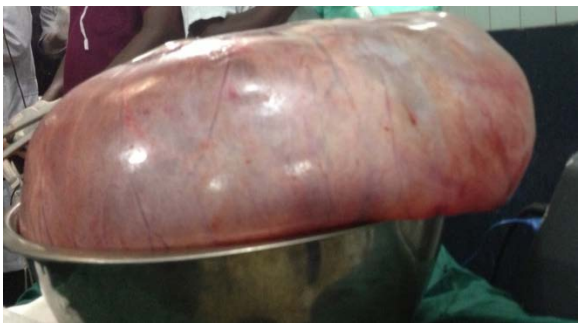

Figure 5: Intact ovarian tumour showing smooth outer surface without external growths.

Right salpingo-oophorectomy was done. A window was created in the ipsilateral broad ligament to access that side of the pelvis retroperitoneum. The corresponding infundibulopelvic and round ligaments were skeletonized. The lesion was removed in one piece through the development of a series of pedicles and ligatures. Haemostasis was secured, and the abdomen was closed in layers. She was transfused with 2 units of whole blood. The right ovarian specimen was sent for histology. The patient had an uneventful recovery and was sent to the post-natal ward.

The baby was received and resuscitated by the neonatologist and sent to the Mother-Baby Unit (MBU). The neonate was given caffeine citrate and intravenous fluids, and breast feeding was initiated within four hours of delivery. The random blood sugar (RBS), heart rate (HR), respiratory rate (RR), S_PO^2 of the neonate were monitored every 2 hours for the first 24-hours. The body temperature of the baby was maintained between 36.5 and 37.5°C

She had an uneventful recovery, and both mother and baby were discharged from the hospital on a post-operative day-5 for scheduled post-natal clinic and then follow-up every 3 months.

Macroscopic and microscopic histopathologic examination reported multilocular ovarian cystic measuring 40 cm at its widest point and weighed 6.8 kg (Figure 6-8). It contained gelatinous substance, had both smooth inner and outer surfaces with no papillae and solid areas. The attached tube was 13 cm long and was grossly normal. On the micro-level, the specimen showed a multilocular ovarian cyst lined by simple mucinous adenomatous cells without stromal invasion, no evidence of pleomorphism or atypia. The picture is compatible with mucinous cystadenoma. The fallopian tube shows insignificant pathological changes.

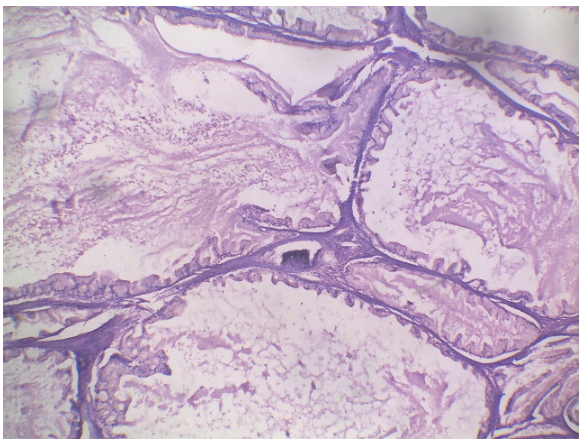

Figure 6: Magnification X40

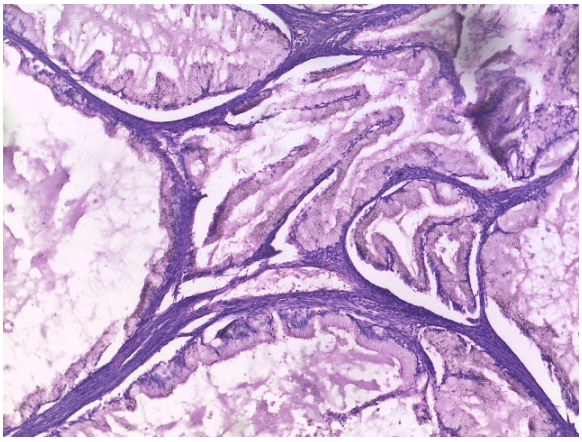

Figure 7: Magnification X 100:

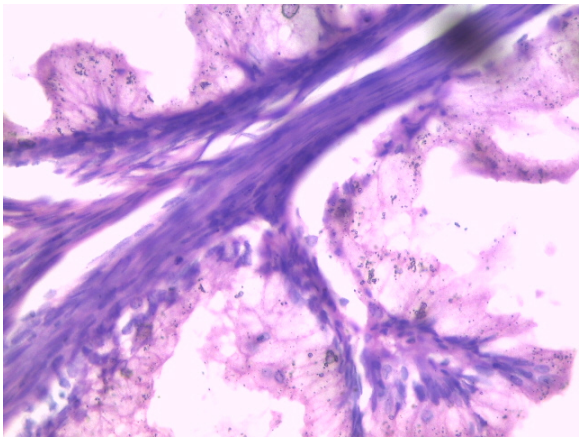

Figure 8: Magnification X 400:

Figure 6,7 & 8: Multilocular mucin-filled cysts supported by scant stroma and lined by bland mucinous columnar epithelium.

Discussion

The availability of sensitive pregnancy test kits, ultrasound scans and increased access and coverage of antenatal clinics in early pregnancy is increasing the detection of asymptomatic and clinically in-apparent adnexal lesions.[11] Accessible and adequate antenatal care, through early detection of danger signs and

management of potential complications, is cardinal in preventing maternal and perinatal morbidity and mortality.[12] An antenatal care in the adolescent age is characterized by poor attendance and inadequate care. Several factors ranging from poor economic support, long commuting distances, through unfriendly care antenatal care programmes to lack of insight into the usefulness of the care are key issues affecting the care. While it is exciting to demonstrate the evidence of pregnancy, the number of foetuses and cardiac activity, it is even more important to be meticulous in ensuring that the adnexae are assessed to rule out masses that may have devastating consequences on the pregnancy.

Ultrasound imaging is important in differentiating between benign and malignant adnexal tumours but difficulties exist in using antenatal ultrasound assessment to differentiate tumours of low malignant potential from benign neoplasms.[13] Certain features like septations, solid areas and papillary projections, when reported on ultrasound scan increase the suspicion for a possible malignant adnexal mass. Although colour flow doppler interrogation may be helpful in the assessment of adnexal masses, it has a false-positive rate of nearly 50% due to the overlap of the low-capacitance blood-flow patterns of pregnancy and malignancy.[14] When in doubt, further evaluation by magnetic resonance imaging (MRI) scan can help to distinguish benign from malignant, with an overall accuracy of 93% for malignancy. [15]

Apart from elevated alpha-fetoprotein level, the rest of the markers: LDH and CA-125, had their serum level within the normal range. The relevance of elevated level alpha fetoprotein was an issue of debate. Some of these oncofoetal molecules are involved in biological function associated with development, differentiation and fetal maturation. Alpha fetoprotein is routinely used for fetal surveillance rather than tumour detection during pregnancy.[16] Elevated maternal serum alpha fetoprotein (MSAFP) may be associated with increased risk of adverse outcomes of pregnancy such as fetal loss, prematurity and intrauterine growth restriction (IUGR).[17] Moreover, placenta and fetal anomalies (morbidly adherent placenta, trisomy 21 and neural tube defect) may also present with an increased level of these markers. The CA-125 is less specific, and its serum level exhibit significant variation in pregnancy.

The findings on evaluation were more suggestive of a benign lesion. Studies recommend surgical intervention for suspected ovarian malignancy, tumour torsion, tumour rupture or obstruction of labour in carefully selected adnexal masses in pregnancy.[3] The size of the mass made ovarian torsion less likely, but complications such as cyst rupture, cyst entrapment and intracystic haemorrhage were anticipated.

The realization of the ideals of ANC is often a difficulty among pregnant adolescents.[12] It is not the usual practice to consider early delivery in the absence of significant obstetric indication. At the time of laparatomy, the caesarean section was appropriate to enhance perinatal outcomes, especially in this case where there were significant socioeconomic challenges. It was not certain that adequate antenatal care would be maintained between the laparotomy and eventual delivery at term, as this was evidenced by the patient's delay in reporting to the teaching hospital. Further, her rural residence and long commuting distance to the teaching hospital made it impractical to delay delivery till term. It is critical that these lesions are also managed by taking cognizance of the specific needs of a marginalized population to optimize maternal and fetal outcomes.

It was, however, difficult to explain the cause of the asphyxia. Birth asphyxia and prematurity are two common causes of neonatal mortality in Ghana.[18] Acute deterioration of fetal heart pattern may have been caused by transient maternal hypotension that may have resulted from aortocaval compression during induction of anaesthesia and the delivery.[19] The presence of a neonatologist for immediate resuscitation and follow up NICU admission were important in ensuring good neonatal outcomes after the delivery.

Although no immunohistochemical staining was done to assess the hormone receptive status of the cyst, earlier case reports have reported rapid growth in mucinous cystadenoma with luteinized stroma.[20,21] These tumours have been reported to demonstrate oestrogen and human chronic gonadotropin positivity on immunohistochemical staining.

Conclusion

Although pregnancy-associated adnexal masses are not uncommon, significant socio-economic challenges may render the diagnosis and treatment of extremely large pelvic masses difficult in pregnant adolescents. The decision to optimize maternal and fetal outcomes is essential in managing rare conditions in marginalized obstetric populations.

Abbreviation

ANC: Antenatal Clinic
MSAFP: Maternal Serum Alpha Fetoprotein
MRI: Magnetic Resonance Imaging
POD: Pouch of Douglas
IUGR: Intrauterine Growth Restriction
LDH: lactic Acid Dehydrogenase
KATH: Komfo Anokye Teaching Hospital

Declarations

Ethics approval and consent to participate

The case report was approved by the Research and Development Unit of the Komfo Anokye Teaching Hospital (KATH). The client was assured of confidentiality. Participation was voluntary and the

patient was informed of the right to pull out at any point which would not affect the care she was receiving. An informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor of this journal.

Consent for Publication

The patient gave a consent for the publication of potentially identifying images and clinical details. The findings and the management were explained to the patient and informed consent sort for publication. The research and development Unit of the Komfo Anokye Teaching also approved the publication of the case report.

Availability of data and materials

The dataset is stored in a repository, this is with the Record Department, Komfo Anokye Teaching Hospital Library, Kumasi. Other details can be made available on reasonable request through the corresponding author.

Competing interests

The authors declare no competing interests.

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