Successful intrauterine aspiration of a large fetal ovarian cyst

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ABSTRACT

A 32-year-old Bahraini lady with a large fetal intra-abdominal cyst detected antenatally on ultrasound examination at 16 weeks of gestation. The cyst was simple anechoic, increasing in size and causing progressive displacement of the fetal thoracic organs. A successful intrauterine needle aspiration was carried out under ultrasound guidance at 30 weeks gestation without maternal or fetal morbidity. Cytology of the cyst fluid showed luteinized granulosa cells and biochemistry demonstrated high concentrations of estradiol, progesterone, and testosterone that confirmed the etiology of the cyst as ovarian. There was no evidence of recurrence following aspiration and no further need for postnatal surgery.

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The first antenatal description of an ovarian cyst was made in 1975 by Valenti et al.[1] Since then, the ultrasound detection of fetal ovarian cyst is increasingly reported in the world literature. There is controversy as to the origin of these cysts, but stimulation of the fetal ovary by both placental and maternal hormones is generally considered to be responsible for this condition in many cases. Complications may occur, however, the most common is probably ovarian torsion which has been observed to occur more frequently during fetal life than postnatally.[2,3] Therefore, to prevent torsion with consequent loss of the ovary, some authors have advocated prenatal ovarian cyst decompression in uncomplicated anechoic cysts. To date, 40 cases of fetal ovarian cysts managed by intra-uterine aspiration have been described.[1,4-6] This is the first case reported from Bahrain and is presented here to highlight its presentation, diagnosis, management and the obstetric outcome.

Case Report. A 32-year-old Bahraini lady was referred to our antenatal clinic following a possible diagnosis of a large fetal intra-abdominal cyst noted on ultrasonography at 16 weeks of gestation in her second pregnancy. Her first pregnancy had ended in a spontaneous vaginal delivery of a healthy female infant weighing 2.81 kg at 40 weeks gestation. There was no significant family or previous medical history. She was O-Rhesus positive with no antibodies. Ultrasound examination was carried out. A large cyst occupying the whole abdomen of the fetus was found (**Figure 1**). This cyst measured 2.7 x 2.5 x 1.6 cm on the transverse and longitudinal scan. The exact origin of the cyst could not be identified. However, the lack of internal echoes within the cyst suggested this was a simple one. The amniotic fluid volume was normal. The fetal kidneys, ureters, urinary bladder and other abdominal organs could not be seen. Amniocentesis was carried out and this resulted in a normal female karyotype (46 xx). Amniotic fluid alpha-fetoprotein level was 12 mg/L and the acetylcholinesterase staining was normal.

The patient was followed by repeated ultrasonographic examination every 2-3 weeks. The

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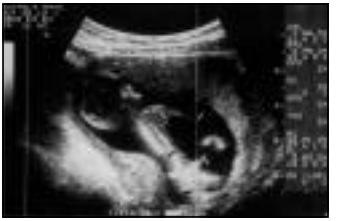

Figure 1 - Ultrasound demonstrating the fetus with a large cyst occupying the whole abdomen.

cyst showed a tendency to increasing in size. At 30 weeks gestation, the cyst measured 10 x 9.8 x 6 cm on the transverse and longitudinal scan. At the same time the fetal organs in the thorax were found to be progressively displaced. In consideration of these findings, fine needle aspiration of the cyst was discussed with the patient and her husband. After informed consent was obtained, the procedure was undertaken on the next day. Under ultrasound guidance a 21 gauge, 15 cm, Sims amniocentesis needle (Cryo Bio System, Paris, France) was inserted transabdominally through the fetal abdomen directly into the cyst and 250 milliliters of a clear yellow fluid was drained. Collapse of the cyst was noted on ultrasound and the cyst was found to be most probably ovarian in origin. Cytologic examination of the fluid revealed few red blood cells, macrophages and a large amount of luteinized granulosa cells. No malignant cells were seen. Fluid endocrinology confirmed the cyst to be ovarian which showed: estradiol 980 000 pmol/L, progesterone 1800 nmol/L, testosterone 75 nmol/L, follicle stimulating hormone 1.8 IU/L, and luteinizing hormone 0.9 IU/L.

After the procedure, the fetus underwent 2 hours of cardiotocographic monitoring, which demonstrated a reactive trace with a complete absence of spontaneous uterine activity. The patient was rescanned every 2 weeks until birth. Follow-up ultrasound examinations showed no more evidence of the cyst, and no fetal abnormalities were detected. Spontaneous delivery occurred at 41 weeks of gestation. A female infant weighing 2.95 kg was born, with Apgar scores of 9 and 10 at 1 and 5 minutes. Physical examination of the neonate was unremarkable. Postnatal ultrasonography was carried out at 2 weeks of age and this confirmed the normal findings with normal appearance of both ovaries, 0.8 cm in length, with no recurrence of the cyst.

Discussion. Various complications are described in association with fetal and neonatal ovarian cysts such as compression on other viscera, rupture of the cyst,[3] hemorrhage,[2,3,7] polyhydramnios,[1,3] ascites,[3] ovarian torsion,[2,3,8] intestinal obstruction,[3] and even sudden infant death.[8] Of these, ovarian torsion is probably the most common. Brandt et al[2] found that in 92% of neonates explored for torsion of an ovarian cyst there was a sonographic evidence that the torsion occurred prenatally. Ovarian torsion can be suspected in utero when there are changes on ultrasound findings from an anechoic cyst to a septated one, a cyst with solid components or a cyst with a fluid-debris level.[2,3] In our case, the ultrasonographic appearance of a thin-walled anechoic cyst suggested this was a simple one. Moreover, the normal appearance of both ovaries on postnatal ultrasound examination suggested that no ovarian torsion had occurred.

Sonographically prenatal determination that the cyst is ovarian is only presumptive. In our patient, elevated levels of estradiol, progesterone and testosterone in the cyst fluid confirmed the ovarian origin. These high steroid concentrations were similar to those reported by others.[4,7] Moreover, the cytology of the cyst fluid in our case showed a large number of granulosa cells. Conversely, low levels of follicle stimulating hormone (FSH) and luteinizing hormone suggest follicular atresia. This discrepancy may reflect the luteotropic effect of placental human chorionic gonadotrophin but does not exclude the possibility of an earlier excessively high FSH concentration that would account for the follicular growth.[9] Therefore, in cases of intrauterine aspiration; cyst fluid analysis may be helpful to confirm the ovarian origin of an intra-abdominal cyst in a female fetus.[7]

After diagnosing the fetal intra-abdominal cyst, 2 main questions arise: (1) should the cyst be aspirated? and if yes (2) what is the preferred time for intervention? Indeed, indications for intrauterine aspiration remain controversial. Although there have been 40 cases of fetal ovarian cyst decompression reported to date, selection criteria for intervention still need to be evaluated. Some authors have limited prenatal aspiration to female fetuses who have a simple anechoic cyst with a diameter of ≥5 cm,[5] while others carried out this procedure even on cases of small fetal ovarian cysts[4] or on those demonstrating "complicated" ultrasonographic appearances.[7] However, in this patient; the cyst was very large (10 x 9.8 x 6 cm) of unknown origin, simple anechoic, increasing in size and causing a marked displacement of the fetal thoracic organs. This gives rise to the question of the best option for the timing of the treatment in this patient. In particular, the question may be debated with regard to the opportunity of postnatal

management. We think, particularly in this case with relatively early gestational age at 30 weeks, waiting until at least the fetal lung to be matured might not prevent possible life-threatening complications especially with progressive organs displacement. Moreover, with this huge cyst that may continue to increasing in size, intrapartum intervention may be necessitated in order to facilitate the delivery or prevent possible birth dystocia. It has been reported that prenatal decompression of a huge abdominal cyst that had markedly displaced the surrounding organs, regardless of its origin, appears to improve fetal survival by preventing associated pulmonary hypoplasia.[10] Therefore, antenatal aspiration was proposed for this patient. Moreover, in cases of fetal ovarian cysts, it has been found that ovarian cyst decompression should be performed in utero to prevent torsion and other complications.[5] Furthermore, there is evidence that in the presence of clinical or sonographic signs of deterioration, as in our patient and previously reported one, prenatal aspiration can be successfully undertaken.[10] This is particularly true when this procedure is carried out in relatively early pregnancy (30 weeks' gestational age), when the fetal abdomen is quite small and the cyst is large enough to prevent movements within the abdomen itself.

On the other hand, several arguments have been addressed against prenatal aspiration. First, ultrasound diagnosis is non-specific, therefore, misdiagnosis may lead to a non-ovarian cyst aspiration. Nevertheless, it seems unlikely that fine needle puncture of structures such as mesenteric cyst, megacystis, or paraovarian cyst will have serious consequences. Furthermore, the favorable outcome in this case and previously described one, demonstrates the value of decompressing the huge intra-abdominal fetal cyst even if its origin is unknown.[10] Such decompression appears to allow the natural progression of the pregnancy and fetal development to continue with avoidance of pulmonary hypoplasia or other possible complications. Second, as with any intra-amniotic procedure, fetal ovarian cyst decompression carries a risk of rupture of membranes, bleeding, infection of the amniotic cavity, and preterm labor. These complications are expected to occur with probably the same frequency as after amniocentesis. Third, needle injuries to fetal abdominal organs such as intestines or vessels may occur. Nevertheless, this complication may be reduced when the cyst is large and lies directly against the fetal abdominal wall. Considering all these reasons, we believe that intrauterine aspiration should be carried out only by well-trained operators who can deal with such cases.

Aside from the criticisms concerning the safety of this procedure, ovarian cyst decompression in utero may be less effective than postnatal aspiration due to its continued hormonal stimulation. However, in our patient and those previously reported, cyst recurrence after decompression has not been observed, even in cases with incomplete cyst aspiration.[1,4-6] Moreover, the risk exists of peritoneal spillage following prenatal aspiration but this is significant only in malignant cases. To date, no case of ovarian malignancy including this patient has been described among fetal ovarian cysts. Furthermore, in ovarian malignancy, ultrasound examination almost always showed a complex mass. For this reason, we concur with others[7] and do not recommend prenatal aspiration in non-anechoic ovarian cysts.

Luckily, in our patient, intrauterine needle aspiration was successfully carried out without maternal or fetal morbidity, with no further evidence of torsion or need for neonatal surgical operation. Therefore, we believe that prenatal decompression of a large fetal ovarian cyst will improve fetal survival by not only preventing pulmonary hypoplasia, but by also reducing the fetal and neonatal incidence of ovarian torsion and its complication and do so with minimal fetal and maternal morbidity. However, further studies are needed to support these results.

References

1. Valenti C, Kassner EG, Yermakov V, Cromb E. Antenatal diagnosis of a fetal ovarian cyst. ***Am J Obstet Gynecol*** 1975; 123: 216-219.
2. Brandt ML, Luks FI, Filiatrault D, Garel L, Desjardins JG, Youssef S. Surgical indications in antenatally diagnosed ovarian cysts. ***J Pediatr Surg*** 1991; 26: 276-282.
3. Nussbaum AR, Sanders RC, Hartman DS, Dudgeon DL, Parmley TH. Neonatal ovarian cysts: sonographic-pathologic correlation. ***Radiology*** 1988; 168: 817-821.
4. Perrotin F, Potin J, Haddad G, Sembely-Taveau C, Lansac J, Body G. Fetal ovarian cysts: a report of three cases managed by intrauterine aspiration. ***Ultrasound Obstet Gynecol*** 2000; 16: 655-659.
5. Bagolan P, Giorlandino C, Nahom A, Bilancioni E, Trucchi A, Gatti C, et al. The management of fetal ovarian cysts. ***J Pediatr Surg*** 2002; 37: 25-30.
6. Heling KS, Chaoui R, Kirchmair F, Stadie S, Bollmann R. Fetal ovarian cysts: prenatal diagnosis, management and postnatal outcome. ***Ultrasound Obstet Gynecol*** 2002; 20: 47-50.
7. Meagher SE, Fisk NM, Boogert A, Russell P. Fetal ovarian cysts: diagnostic and therapeutic role for intrauterine aspiration. ***Fetal Diagn Ther*** 1993; 8: 195-199.
8. Kasian GF, Taylor BW, Sugarman RG, Nyssen JN. Ovarian torsion related to sudden infant death. ***Can Med Assoc J*** 1986; 135: 1373.
9. Montag TW, Auletta FJ, Gibson M. Neonatal ovarian cyst: prenatal diagnosis and analysis of the cyst fluid. ***Obstet Gynecol*** 1983; 61: 38S-41S.
10. Ito M, Yoshimura K, Toyoda N, Tanaka H. Aspiration of giant hepatic cyst in the fetus in utero. ***Fetal Diag Ther*** 1997; 12: 221-225.