Fetal ovarian cyst with prenatal torsion of the pedicle diagnosed in the third trimester: A case report

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**ABSTRACT**

Ovarian cysts develop rarely in fetuses during pregnancy and usually disappear after birth. However, during pregnancy, torsion and rupture of the cyst can occur and it is necessary to manage such cases. At present, there is no standardized prenatal or neonatal treatment. However, the preservation of ovarian function is an important consideration. Here, we present a case involving a 35-year-old woman who gave birth to an infant with a complicated ovarian cyst, which was resolved through laparoscopic surgery on the third day after birth. The prenatal diagnosis of the fetal ovarian cyst was performed prenatally using ultrasound and magnetic resonance imaging (MRI). However, torsion of the ovarian pedicle was suspected because of dorsal thickening of the cyst wall and fluid formation indicated by high intensity on T1- and T2-weighted MRI. Surgery was complicated due to involvement in the torqued pedicle of the left fallopian tubal fimbria, which was released. The cyst was drained and partially resected, and then the pedicle torsion was released. By four years and five months of follow-up, there had been no ovarian cyst findings on ultrasound or MRI. Larger cysts are more likely to involve torsion of the ovarian pedicle and potential bleeding. Considering that, in this case, the cyst was large and proved to be ischemic–hemorrhagic, the decision to operate seemed justified.

1. Introduction  

Ovarian cysts are the most common abdominal anomalies in fetuses, occurring in approximately one per 2000–3000 deliveries [1]. Maternal diabetes mellitus, pregnancy rhesus factor (Rh)-incompatible, and fetal hypothyroidism can increase fetal gonadotropin levels, leading to the formation of fetal ovarian cysts [2]. These cysts can develop during pregnancy; most are believed to disappear after birth. During the course of pregnancy, torsion of the pedicle and rupture of the ovarian cyst may occur, which are important to manage. However, no definite treatment plan exists for antenatal and neonatal ovarian cysts. The goal of treatment is to preserve ovarian function as much as possible [3]. Here, we report a case of fetal ovarian cyst with prenatal torsion of the pedicle.

2. Case Presentation  

A 35-year-old woman, para 1–0–0–1, had a spontaneous pregnancy and no problems on prenatal checkup. At 34 weeks of gestation, the patient was referred for examination of a fetal intra-abdominal cyst. Ultrasound revealed a large (5-cm) cyst in the lower abdomen of the fetus. The cyst was fluid-filled, without any solid component (Fig. 1). Suggested differential diagnoses included ovarian cyst stalk torsion, duplicated intestinal tract, and hydronephrosis. There were no abnormalities, such as pleural effusion or ascites. The external genitalia were normal. T2-weighted magnetic resonance imaging (MRI) showed a 5-cm cystic mass on the right side of the upper fetal pelvis (Fig. 2), as had been seen on ultrasonography. Both T1- and T2-weighted images showed high signal intensity inside the mass. Fluid formation and thickening of the cyst wall on the dorsal side indicated intracystic hemorrhage, which suggested an ovarian cyst with torsion of the pedicle.

At 37 weeks and four days of gestation, a female infant was delivered by cesarean section (because of repeated cesarean section). The birth weight was 3208 g, the Apgar score was 8/9, and the cord pH was 7.239. The infant's general condition was good on the second day after birth. However, neonatal T2-weighted MRI showed a suspected right ovarian cyst with torsion of the pedicle (Fig. 3). In order to preserve the child's ovarian function, it was decided to surgically release the torsion. Laparoscopic surgery under general anesthesia was performed on the
following day. Intraoperative findings showed a right ovarian cyst with torsion of the pedicle. The left fallopian tubal fimbria was caught in and adhered to the torsion site of the right ovarian cyst, which made it difficult to release. The twisted right ovary appeared to be ischemic–hemorrhagic (Fig. 4). Partial resection of the right ovarian cyst and adhesiolysis of the left fallopian tube were performed. Approximately 50 mL of dark reddish brown cyst fluid was removed; the torsion was released. The right ovarian cyst was diagnosed pathologically as serous cystadenoma with ciliated epithelium and mild cell vacuolation.

At last follow-up, when child was aged 4 years and 11 months, there were no ovarian cyst findings on ultrasound or MRI.

3. Discussion

Differential diagnoses of fetal cystic pelvic masses include ovarian cyst, duplicated intestinal tract, and hydroureter, among others. A study by the Canadian Consortium for Research in Pediatric Surgery suggested factors that differentiate fetal ovarian cysts from non-ovarian cysts. A clear organ of origin, diagnosis earlier in gestation (23.5 weeks vs. 33.5 weeks), smaller initial prenatal cyst diameter (15.8 mm vs. 39.7 mm), and sonographic cyst character change (simple to complex) are associated with non-ovarian origin [4]. In the reported case, the fetal cyst was diagnosed late in gestation as a simple 5-cm cyst; hence, the diagnosis of ovarian cyst was made.

However, an ovarian cyst with torsion of the pedicle was suspected and there was concern to preserve ovarian function. Although the benefits of aspiration of fetal cysts have not been proven, aspiration has been described in the management of larger, presumed ovarian cysts. An open randomized controlled trial reported that in utero aspiration of anechoic fetal ovarian cysts did not lead to a reduction in overall neonatal interventions [5]. However, a systematic review and meta-analysis showed that the overall spontaneous resolution rate of conservatively managed cysts was 46%; the rate of prenatal torsion in simple cysts <40 mm in size was lower in aspirated cysts than in conservatively managed cysts [6]. A change in the ultrasound pattern of a cyst is associated with an increased risk of ovarian loss [7]. In the reported case, fluid formation in the fetal cyst, and the thickened cyst wall, led to a diagnosis of bleeding due to torsion of the pedicle. Antenatal aspiration might have been chosen as a treatment.

There is no standard treatment for an ovarian cyst in a neonate. Many reports use the size of the cyst as a criterion for neonatal treatment [8]. For asymptomatic girls, families are counseled regarding observation versus diagnostic laparoscopy. For large (≥4 mm) or complex cysts, operation may be favored. In theory, ovarian torsion may occur during periods of observation. A necrotic, torqued ovary has been reported to cause bowel obstruction and urinary tract obstruction in infants. The concept of early laparoscopic exploration is supported by case series in which early operation was associated with high rates of adnexal preservation. However, a review by Brandt et al. reported that ultrasound evidence of torsion was found in 92% of surgical cases at or before birth, which suggests that most torsion occurs prenatally; the risk of postnatal torsion may be low [9]. There are more cases of torsion in which the size of the cyst exceeds 40–50 mm, although many cases resolve spontaneously. Complex cysts and those of size 40 mm or more are related to ovarian torsion and the performance of postnatal surgery [10]. In the reported case, the neonate’s general condition was good but neonatal MRI revealed a right ovarian cyst with torsion of the pedicle. Laparoscopic surgery was performed to release the torsion and preserve ovarian function.

During surgery, torsion of the pedicle was observed but was difficult to release because of strong capture and adhesions. Worsening of the tortuosity was prevented and the normal fallopian tube tissue was preserved by opening the window. Thus, the normal fallopian tube tissue may have been preserved. Long-term follow-up data indicates that, although the twisted adnexa appears to be ischemic–hemorrhagic, it can
safely be restored by detorsion, with the preservation of ovarian function [11]. In the reported case, recovery of ovarian function can therefore be expected. Long-term follow-up is planned.

Contributors

Hayase Nitta drafted, reviewed and edited the manuscript, and provided patient care.

Tadatsugu Kinjo reviewed and edited the manuscript, and provided patient care.

Yoshino Kinjyo reviewed and edited the manuscript, and provided patient care.

Hisako Yamada reviewed and edited the manuscript, and provided patient care.

Hitoshi Masamoto reviewed and edited the manuscript, and provided patient care.

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