Torsion of adnexal cyst in utero – case report

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Abstract

Background: Fetal tumors represent rare group of abnormalities however ovarian cysts are the most frequent type of abdominal tumors in female newborns. The diagnosis has become more frequent due to the prenatal ultrasonography monitoring. Due to the possible complications of such abnormalities it is important to be aware of the management and treatment options. The literature presents different management options from conservative approach such as wait and see policy to more aggressive as intrauterine cyst aspiration or surgery. Case report: We have presented the management and treatment of large complex fetal ovarian cyst diagnosed prenatally. Towards the end of pregnancy the cyst changed its consistency; it contained fluid/debris levels which did show some calcifications and possible hemorrhagic fluid suggesting torsion. The patient was delivered by a cesarean section and the neonate was referred for selective abdominal surgery. Conclusion: Fetal ovarian cysts are rare however they may cause serious complications. The decision regarding the management and treatment of fetal ovarian cyst should depend on the size, appearance of the cyst and visible complications. Ultrasonographic monitoring allows differentiation of the ovarian mass (simple/complex) and application of appropriate management. We think that simple ovarian cyst should be managed by wait and see policy and close postnatal follow-up and complex ovarian cysts should be managed by early surgery especially if symptomatic.

Key words: fetal ovaria cyst

Introduction

Fetal tumors represent rare group of abnormalities however ovarian cysts are the most frequent type of abdominal tumors in female newborns [1]. The rates of detection of fetal ovarian cysts have increased since the advent of routine antenatal sonography. The first case of an ovarian cyst was reported in 1889 in a stillborn premature whereas the first successfully treated ovarian cyst in newborn period was done by Bulfamonte in 1942 [2]. The etiology of fetal ovarian cysts has not been entirely clarified. Most likely, it results from fetal exposure to maternal and fetal gonadotrophins, found often in the mothers having increased levels of beta-HCG (diabetes, Rh isoimmunization and toxemia). Other suspected hypotheses include fetal immaturity, fetal hypothyroidism as well as placental insufficiency in addition to incomplete maturation of the gonadostat [3]. Various complications are described in association with ovarian cysts: compression of other viscera, rupture of the cyst, hemorrhage but the most common is ovarian torsion with possible consequent loss of the ovary. According to Nussbaum’s classification, ovarian cysts can be divided into simple (completely anechoic) and complex (characterized by fluid-debris level, clot, septa, and echogenic wall) cysts suggesting torsion [4]. There is still considerable controversy regarding treatment of ovarian cysts. The literature presents different management options such as wait and see policy, aspiration or surgery. In this case we present the management as well as successful treatment of large complex neonatal ovarian cyst diagnosed antenatally.

Case presentation

A 34 year-old mother at 33 week of gestation, gravida 2, with a history of spontaneous abortion was admitted to the Department of Perinatology and Gynecology because of lower abdominal pain and suspicion of fetal ovarian cyst. Fetal ultrasound had shown a 67 × 43 mm simple anechoic cyst at the right side of fetal pelvis (Fig. 1, 2). The otherwise normal anatomy of the gastrointestinal and urinary tracts in a female fetus was highly suggestive of an ovarian cyst. The otherwise normal anatomy of the gastrointestinal and urinary tracts in a female fetus was highly suggestive of an ovarian cyst. Otherwise the fetal anatomy was within the norms. All the routine hematological and biochemical investigations were within normal limits. After diagnosis the mother was followed by serial ultrasound scans every 2 weeks to check for the sonographic disappearance of the cyst or for possible shift to the pattern of torsion. The scan at 36 weeks re-
revealed different echogenicity of the ovarian cyst and possible change to complex type of the cyst (Fig. 3).

Fig. 1. Fetal ultrasound shows a 67 × 43 mm simple anechoic cyst at the right side of fetal pelvis at 33 weeks

Fig. 2. The fetal abdominal cyst in 3D scan

Fig. 3. The hyperechogenic complex ovarian cyst at 36 weeks reflecting the change of cyst character

The diameter was 65 mm. Following, at 39 weeks the patient was admitted to the hospital for elective cesarean section, however at that time the ultrasound scan of the ovary showed shift to echogenic complex ovarian cyst (fluid/debris level) (Fig. 4, 5).

Fig. 4. The ultrasound scan of the ovary shows a shift to echogenic complex ovarian cyst (fluid/debris level) at 39 weeks

The cyst did not change in size however there was change of the consistency and echogenicity. It contained fluid/debris levels which did show some calcifications and possible hemorrhagic fluid. The patient underwent cesarean section at 40 week of gestation and delivered female newborn weighing 3770 g and Apgar score 10, 10.

Fig. 5. The ovarian cyst in 3D scan at 39 weeks. The complex cyst with intracystic septa, fluid/debris level

The fetus was evaluated by neonatologist who confirmed good neonatal state and the presence of ovarian cyst without any other abnormalities of anatomical structures. The only disturbing abnormality detected was increased level of AFP (26138 ng/ml) and CEA (30.5 ng/ml). The patient was transported to the Department of Pediatric Surgery 6 days after delivery for selective abdominal surgery. Laparoscopic exploration revealed a large dark brown mass localized in right abdominal compartment extending from subhepatic region to the pelvis. After evacuation of a dense hemorrhage fluid from the cyst the torsion of right ovary secondary to the large ovarian cyst was found as a cause of pathology. Right salpingo-oophorectomy was performed because
necrosis made the distinction of normal ovarian tissue impossible. After excision of the adnexa there were two solid adhesions between right ovary and the wall of jejunum, which were excised and the wall of the jejunum was checked. The histopathology examination indicated the presence of hemorrhagic necrosis and foci of dystrophic calcifications. With these features, a diagnosis of a congenital ovarian cyst with torsion was given. The postoperative course of the patient was uneventful and neonate was discharged home on the 3rd postoperative day. The ambulatory follow-up indicated normal physical development without additional problems.

Discussion

The incidence of ovarian cysts has been estimated at more than 30% (this estimate is based on an investigation of stillborns or infants who died within 28 days after birth) [5]. Ovarian cysts arise from mature follicles which are usually < 2 cm in diameter, cysts larger than that size are considered pathological which can be diagnosed usually beyond 28 weeks. The diagnosis has become more frequent due to the prenatal ultrasonography monitoring. Torsion is the most common complication (50-70%) and it has been observed to occur more frequently during fetal life than postnatal. Some studies have reported that the outcome of such complication is related to the length of the cyst pedicle which is more predictive than cyst dimension [6], however unfortunately it may only be evaluated during surgery. Complication such as torsion may cause inflammatory adhesions which were observed in case of our patient and probably induced the increase in the AFP and CEA level. Other complications may include intracystic hemorrhage, rupture, less frequently urinary tract or bowel obstruction and rarely autoamputation of the cyst. However the management of ovarian cyst should be based on the size and echopattern of the cyst. Few authors have shown that unilateral simple cyst is suggestive of benign process. It is known that cyst < 4 cm have a tendency for spontaneous resolution both prenatally and after delivery which means that they can be managed conservatively by serial scans. Bagolan et al. managed 34 simple ovarian cyst < 5 cm with wait and see policy and they have observed spontaneous regression in 26 cases, 1 persistence at birth and 7 cases of torsion [10]. Others have reported that simple cysts even up to 8 cm can safely be monitored by ultrasound, as spontaneous resolution can occur within 4-5 months [4, 15]. That is why we think that the most appropriate clinical approach in case of simple ovarian cysts is to adopt a wait and see policy which requires periodic ultrasound monitoring to assess the course of the condition. In our patient the diagnosis was made at 33 weeks of pregnancy, before that the patient was monitored regularly by ultrasonography. Based on the regular US scans for few weeks we were observing a large simple ovarian cyst that is why we decided to proceed with the wait and see policy.

Another management option presented by few studies is intrauterine ovarian-cyst aspiration [9, 11]. Bagolan et al. in their study performed in utero aspiration of 14 simple cysts measuring ≥ 5 cm observing resolution in 12 cases and torsion in 2 concluding that “in utero” aspiration of ovarian cyst is a safe procedure. They have also recommended surgical management for cysts with ultrasound pattern of torsion persisting postnataley [10]. However others think that continued hormonal stimulation leads to regrowth of the cyst [12], can lead to peritonitis due to rupture or spillage of cyst fluid but most importantly the increased possibility of wrong diagnosis. US scan cannot distinguish benign and malignant lesion especially in the presence of complex mass. There have been reports of cystoadenoma and bilateral granulosa-cell carcinomas of the ovary have been reported in a 30-week-old fetus [7, 8] Because we weren’t certain about the type of the ovarian mass we did not want to proceed with intrauterine aspiration.

Another question which has to be asked is what is the best time for delivery in case of cyst with the ultrasound pattern of torsion. In decision making we have to consider maturity of the fetus, possible attempts to save the ovary and possible complications. We think that due to the possibility of ultrasound monitoring we can observe the ovary on day time bases and be able to make the decision according to the changes that we observe. Since there was no change in the size of the ovary and we were not sure if the ultrasonographic evidence of complex cyst is actually a torsion we decided not to proceed with early delivery. The patient was delivered as soon as we were sure that the fetus obtained pulmonary maturity. According to the literature early delivery is not currently recommended.

What to do after delivery? Due to the fact that ultrasound is nonspecific when it comes to interpreting the nature of fetal ovarian cyst and does not give us a sure answer about the possibility of torsion we should take a moment to think about treatment option. The possibilities include wait and see policy, laparoscopy explorations and laparotomy. According to the study by Kwak et al., postnatal symptomatic cysts or cysts with diameter greater than 5 cm that do not regress or enlarge should
be treated, but uncomplicated asymptomatic cysts less than 5 cm in diameter should only be observed and re-assessed by serial ultrasonography [13]. Whereas, Kessler et al. recommended US-guided aspiration of asymptomatic large ovarian cysts for salvage or for decompression if torsion occurs. Surgery was to be reserved for patients with acute torsion, intestinal obstruction and intestinal volvulus [14]. Sometimes such a choice of treatment could be dangerous even with lethal demise [15]. Monnery-Noche et al. found that most of complex appearing ovarian cysts were torsed. Because ultrasonography was not able to distinguish torsed from hemorrhagic cyst the early surgery was recommended to define diagnosis and assessment of ovarian viability [16]. In our case we have decided to proceed with surgery because we were unsure about the character of ovarian cyst, we wanted to find out if there is any functional ovarian tissue left as well as visualize other structures which could be involved in the process.

To conclude the decision regarding the management and treatment of fetal ovarian cyst should depend on the size, appearance of the cyst and visible complications. Ultrasonographic monitoring allows differentiation of the ovarian mass (simple/complex) and application of appropriate management. We think that simple ovarian cyst should be managed by wait and see policy and close postnatal follow-up and complex ovarian cysts should be managed by early surgery especially if symptomatic.

References