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Ovarian torsion due to prenatally diagnosed cystic abdominal mass – case report and literature review

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ABSTRACT

The majority of abdominal cystic formations in newborns are benign and usually originate from the urinary tract, genital system or gastrointestinal system. Fetal ovaries are predisposed to development of cysts due to the maternal hormonal environment of advancing gestational age. Prenatal diagnosis became possible with improvement of ultrasound techniques. Complex pattern of the cysts are at higher risk for complications, the most concerning being ovarian torsion and consequent compromised fertility. Management is controversial, involution of simple cysts few months after birth is possible considering the cease of ovarian stimulation by maternal and fetal hormones but surgical approach is more indicated in symptomatic or complex cysts. We report the case of a prenatally diagnosed cystic abdominal mass at 33 weeks of gestation with antenatal ovarian torsion and need for right oophorectomy in the neonatal period. At two months of age appearance of ovarian cyst was observed on the contralateral ovary by ultrasound, being managed conservatively.

Keywords: abdominal mass, neonatal ovarian torsion, fetal ovarian cyst

Abbreviations:
MRI – magnetic resonance imaging
hCG – human chorionic gonadotropin
FSH – follicle-stimulating hormone

LH – luteinizing hormone

INTRODUCTION

The majority of the fetal-neonatal abdominal masses are benign, genital localization being the second most common appearance in female fetuses, followed by renal or gastrointestinal tumorous masses [1]. The burden of fetal ovarian cysts is relatively frequent, incidence being approximately 1 to 2500 live births and is mainly the result of a rich hormonal environment: maternal estrogens and human chorionic gonadotropins stimulate the fetal ovary, causing fetal ovarian follicle production and formation of ovarian cysts [2-4,6]. Hence, cysts tend to appear in the third trimester due to high hormonal levels and can present spontaneous involution few months after birth due to cease of stimulating hormones. Other endocrine causes are also mentioned to cause fetal-neonatal ovarian cysts: immaturity of gonadotropin secretion, fetal hypothyroidism, congenital adrenal hyperplasia [8]. Despite of the natural history of spontaneous regression of the cysts, in some cases they still can continue to grow on behalf of increasing FSH-LH levels in infants in the first three months of life [5,8,12].

Majority of the cysts are unilateral and associated anomalies are rare [2,5]. Incidence is also increased by several maternal factors like diabetes mellitus, preeclampsia, Rh isoimmunization due to greater hCG levels because of larger placenta in complicated pregnancies [6,8].

Fetal-neonatal ovarian(?) cysts can be classified into two main types based on their size and appearance. Simple or physiologic cysts have a diameter less than 2 cm with clear fluid, are usually asymptomatic and show a higher tendency for spontaneous involution [7,8,11]. Large or complex cysts are usually greater than 2 cm, can contain detritus, septs, solid components, coagulated blood fragments or have echogenic walls with a probability of ovarian-vascular dysgenesis [7,8,11,19].

Small cysts can be asymptomatic, but they can also cause pain, irritability, vomiting, abdominal destintion [8]. The most frequent complication is represented by ovarian torsion, especially in case of larger cysts which can lead to short and long term complications as hemoperitoneum, ascites, intestinal obstruction, autoamputation of the ovary and compromised fertility [8]. Torsion occures more frequently on the right side [14]. Anemia can appear due to intracystic hemmorrhage. Obstruction of gastrointestinal or urinary tract, respiratory distress due to mass effect on diaphragm, difficult delivery as a result of abdominal dystocia all represent possible complications [8-10].

Exact localization of the mass is useful in deciding the belonging of the tumor and thus distinghuishing a series of other possible conditions: reproductive tract anomalies, urinary tract obstructions, urachal cyst; mesenteric or omental cyst, volvulus, colonic atresia, duodenal atresia, intestinal duplication cyst, biliary cyst, pancreatic cyst; cystic meconium peritonitis; splenic cyst, lymphangioma; beningn cystic teratoma [7,8].

There is no general agreement in management of these cysts in fetus or neonate. The spontaneous involution is weighed against the possibility of complications, especially ovarian torsion [7]. Prenatal management is based on monitoring the cyst regarding to size, character and potential complications. Prenatal aspiration of large cyst is possible, but with a series of complications [7]. After birth an ultrasound examination should be performed, followed by repeating the scan every 4 to 6 weeks until resolution. Aspiration or surgical removal of the cyst are also accepted options especially in case of amplification in size, associated signs of torsion or persistance above 4 months [7,8].

CASE REPORT

Patient history

A 30 year old woman, gesta II, para I, was diagnosed with a fetal abdominal cystic tumor at 33 weeks gestational age. Tendency of tumor growth was observed until 35 weeks gestational age, when stabilizing was observed. The pregnancy evolved normally. The past medical history and antenatal course was unremarkable. Birth was achieved by cesarean section due to failure to progress in labor, hypotonic and hypokinetic labor, risk of uterine rupture. Ascending maternal infectious risk was present since membranes were ruptured 45 hours prior to birth, amniotic fluid was bloody. A female infant was delivered weighting 3660 g, length was 54 cm, head circumference was 34 cm. Apgar scores were 9 at 1 minute and 10 at 5 minutes and the newborn showed a good postpartum adaptation.

Clinical status and initial imaging findings

The infant's physical examination showed pathology and vital signs were stable. After birth abdominal ultrasound revealed a well-defined complex cystic mass in the lower abdomen with measurements of approximately 4.72/3.98 cm. (Figure 1 and 2) In order to get a more detailed image of the mass, abdominal MRI was performed showing an abdominal tumorous mass with intraperitoneal localization at the level of the right colic flexure, with measurements of 43/35 mm, high protein content. Suspicion of peritoneal cyst or intestinal duplication cyst was raised, but neither the abdominal ultrasound, nor the MRI could specify the exact location of the mass and whether the tumor belonged to the gastrointestinal or genitourinary system.

Alpha-fetoprotein was measured for possible germ cell tumor but showed normal values. Hematological findings showed leukocytosis (28.150/mm^3) with elevated procalcitonin (4.17 ng/dL; normal value <0.5 ng/dL). Along with the prolonged rupture of membranes, laboratory findings were interpreted as a neonatal infection, therefore empirical associated antibiotic therapy with ampicillin/sulbactam and amikacin was carried out for 5 days.

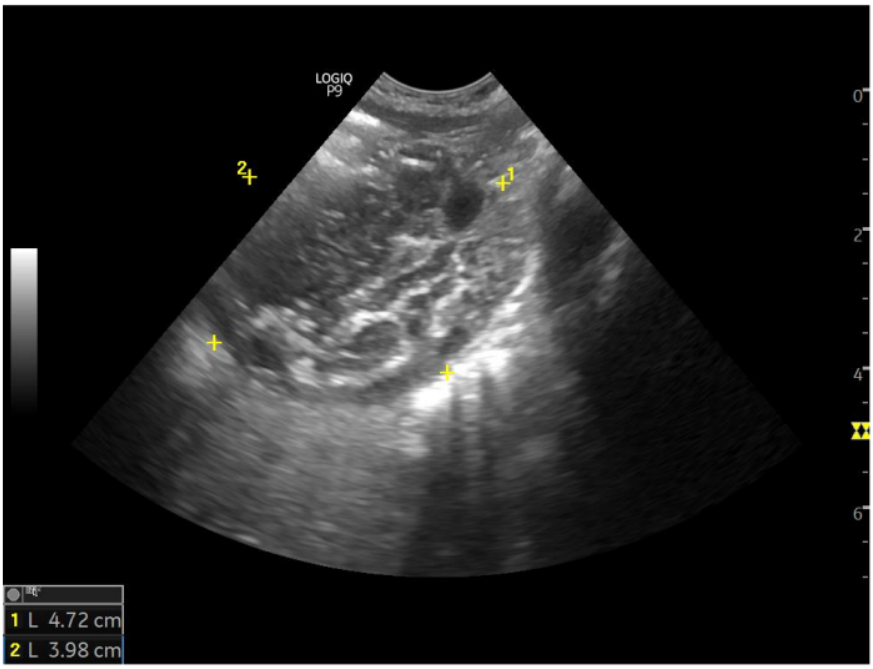

Figure 1: Ultrasound image of the abdominal mass in the first day of life – heterogenous solid compotent in the right ovary, with fluid-filled cysts

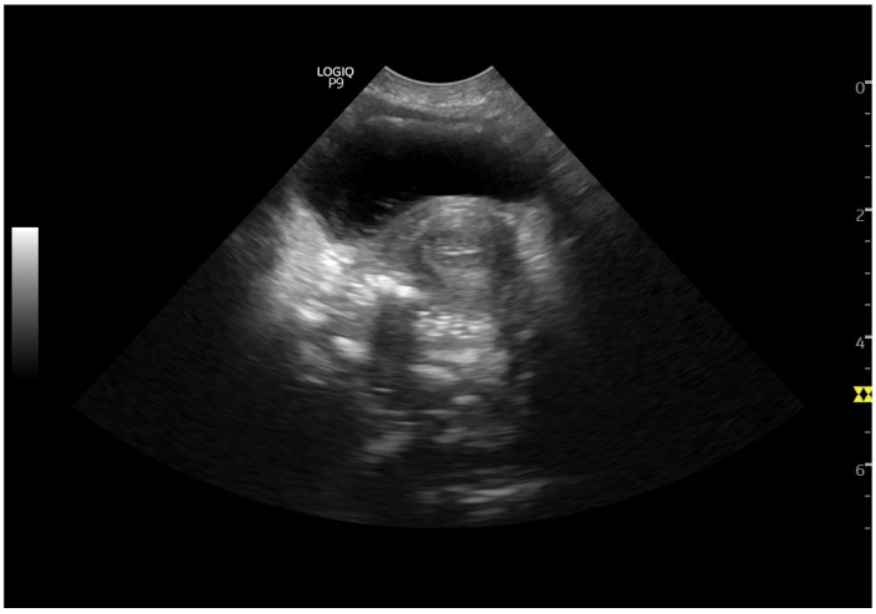

Figure 2: Ultrasound image of the abdominal mass in the first day of life – fluid-debris level and echogenic areas

Pediatric surgeons raised the suspicion of an intestinal duplication cyst and recommended exploratory laparotomy for diagnosis and surgical repair but the mother decided to discharge the infant in order to get a second opinion.

One day after discharge, at 16 days of life the infant was brought to the pediatric emergency department for bloody stools and was eventually admitted to Pediatric Surgery and Orthopedics Clinical Department of Targu Mures. At 17 days of life exploratory laparotomy was performed which showed a necrotized right ovary possibly due to antenatal torsion. (Figure 3) Intraoperative gynecologic consult was requested and exploratory laparotomy was converted to classical approach in order to perform right oophorectomy. Postoperative course was uncomplicated, discharge being possible 4 days after surgery in good general condition; stable cardiorespiratory status; soft abdomen; without tenderness; passage of normal stool; normal urine output.

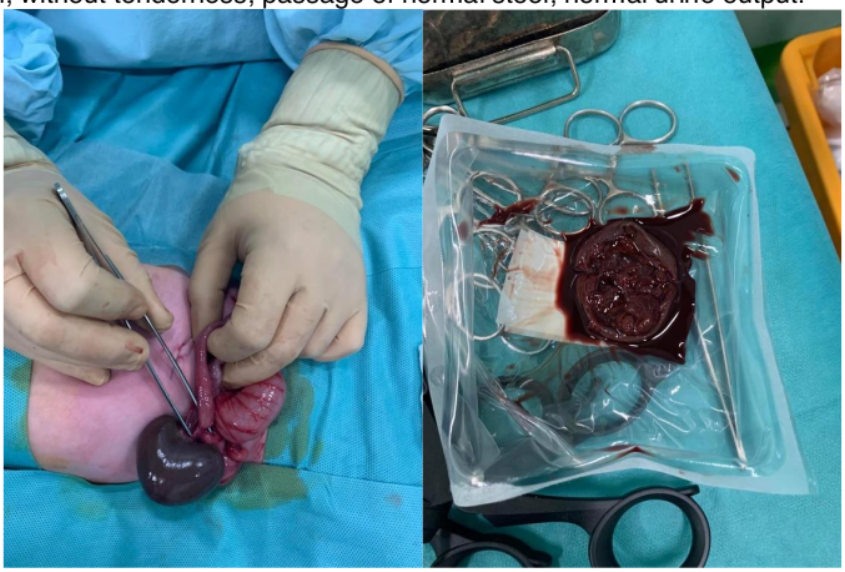

Figure 3: Intraoperative appearance of the right ovary

Histopathological examination revealed hemorrhagic infarction of the right ovary. Macroscopic examination described a tissue fragment of 40x34x22 mm with a smooth,

dark purplish color and homogeneous appearance, one section characterized by blood clots. All sections presented a blurry microscopic view, primordial follicles and fimbriae structures were seen accompanied by hemorrhagic infarction and mass extravasation of erythrocytes. The ovarian cortex was characterized by multiple calcified areas and thrombosed vessels along with areas filled with hemosiderin loaded macrophages.

Follow-up and monitoring

Two weeks post-operative follow-up was unremarkable. At 2 months of age the infant is admitted to territorial hospital with acute bronchiolitis, during hospitalization she starts presenting bloody stools. Abdominal ultrasound raises the suspicion of left ovarian torsion hence she is transferred to Pediatric Surgery and Orthopedics Clinical Department of Târgu Mureș. Native abdominal CT describes the followings: mild intestinal distension; lack of free intraperitoneal fluid; isodense, homogeneous mass with well-defined edges, possibly at the left axial lodge with measurements of 22/20 mm.

Abdominal ultrasound is repeated by gynecologist and pediatric specialists describing a well-circumscribed inhomogeneous mass of approximately 3.30/1.54 cm, with hipo- and hiperechogenous areas, positive Doppler sign, localization in the left iliac fossa, unable to say the belonging to organ. (Figure 4) We mention the fact that no cystic mass was observed on the left side by any of the imagistic studies in the neonatal period, but during the exploratory laparotomy, the left ovary showed uneven surface with small but visible possible cysts.

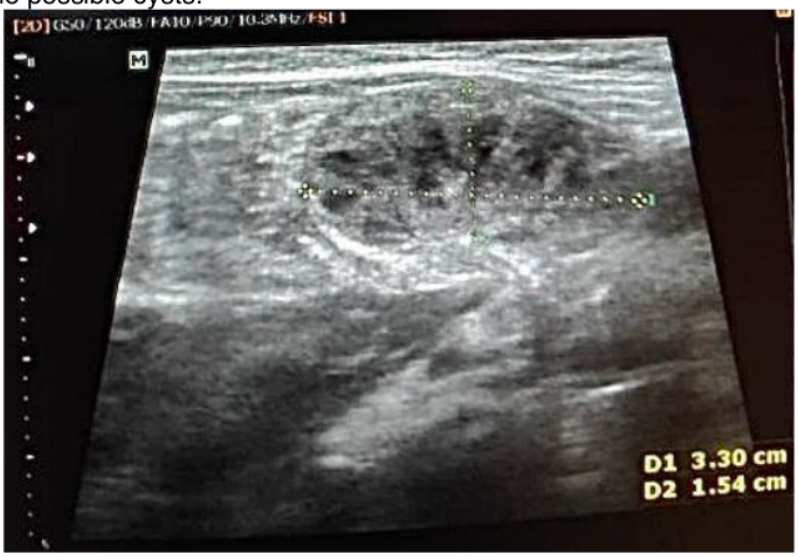

Figure 4: Ultrasound image of the left ovary at 2 months of age

Given the good clinical status, multidisciplinary team by radiologist, anesthesiologist and gynecologist decides postponing the abdominal MRI and maintain further conservative approach by abdominal ultrasound follow-up.

Regular follow-up by adominal ultrasounds was carried out with gradual decrease of the cyst in size. The patient is now a healthy two years old little girl. Last ultrasound at two years old revealed a left ovary with approximately 12 mm lenght in long axis, with small 2-3 mm follicles.

DISCUSSION

Most of the abdominal cystic masses in female fetuses originate from the ovaries. Timing of diagnosis is usually the end of the second trimester or in the third trimester. In the study by *Akın et al.* 90% of the patient were diagnosed antenatally, time of

diagnosis between 34-38 weeks, and about 75% of the patients had unilateral right cyst [8]. Furthermore, pregnancy complications like gestational diabetes, preeclampsia, polyhydramnios, Rh incompatibility were present in about one-third of the patients. Our diagnosis of fetal abdominal cystic mass was established at 33 weeks, with a slight increase in size until 35 weeks and stagnant evolution afterwards. No other pregnancy related problem was noted before or after this, lacking any maternal risk factors for fetal abdominal mass, moreover the first pregnancy went unremarkable delivering a healthy female newborn at term.

Malignancy is very rare among fetal-neonatal ovarian cyst, so the interest is primarly focused on the possible complications caused by the cysts. Ovarian torsion is more common in case of cysts larger than 5 cm and occurs preponderant on the right size because of the anatomical position of the sigmoid colon with the left ovary, which was also the case of our patient [14,15]. Most retrospective studies observed that torsion occurs antenatally and only in rare cases after birth [8,13,21]. Signs of torsion included simple cysts changing its character by transforming into complex cyst, intracystic hemorrhage, no change in direction of involution, followed by further complications such as hemoperitoneum, adhesion to other organs and obstruction, calcification of the cyst [8]. Based to the study of *Özcan et al.* and *Devriendt et al.*, lack of vascularization on Doppler ultrasound is not a pathognomonic sign for torsion, but fluid-debris level was seen in majority of their patients with ovarian torsion [13,22]. Absence of flow can be related to inexperience of sonographer or young age of the patient [17,20]. The ovaries have a rich blood supply by the paired ovarian arteries, therefore a partial torsion can present with a normal arterial flow. *Prieto et al.* reported that only 38% of torsioned ovaries were detected by absence of Doppler flow [17].

According to *Nussbaum et al.*, our patient had a complex cyst on the right ovary with suggestive factors for torsion. Giving the antenatal history of prolonged rupture of membranes, neonatal infection can be questionable, systemic inflammatory markers may have been raised also because of the torsion. Leukocytosis could be the result of cortisol release due to ischemia [16].

Before birth, observation by ultrasound is recommended. Although prenatal ultrasound-guided aspiration of cysts may reduce the risk of complications and preserve fertility, prenatal invasive procedures can have a great impact on the fetus [7,18]. Postnatal management is controversial because most patients are asymptomatic at birth, spontaeous regression is possible but hormonal cause is not the only ethiopathogenic factor and compromised fertility because of ovarian torsion is a serious unwanted complication [19]. *Enríquez et al.* compared the management of a retrospective surgical group with a prospective conservative group and reported the involution of 11 cysts within 1 year, therefore underlined the importance of follow-up and preservation of the ovary if possible, even in case of complex cysts [19]. *Chiaramonte et al.* also reported a regression of complex cyst within 1 year in half of the cases [14]. If surgery is indicated, cystectomy should be tried in the first place, in order to preserve the ovary. Even though cysts were not seen by ultrasound on the left side, their detection at two months old and after suggests bilateral ovarian cysts. It is possible that torsion occured on the right side because of larger cysts and due to anatomical localization. Cysts on the left side were not detected in the neonatal period by imagistics, but were seen during surgery. They were first evidenced by ultrasound at two months of age, possibbly because they showed a temporary growth during infancy period. Left ovarian cyst were managed conservatively and showed a slow regression at two years old.

CONCLUSION

We presented a case of a newborn with bilateral ovarian cysts; the right ovary being torsioned antenatally and treated surgically and the left ovary being managed conservatively. Once detected, serial ultrasounds are decisive in management of these

cysts. Fluid-debris level can indicate an antenatal ovarian torsion. All measures should be made in order to preserve maximal ovarian parenchyma.

PATIENT CONSENT

The minor's legal guardian provided a written consent for the publication of this case report.

CONFLICT OF INTEREST

The authors have no conflict of interest to declare. All co-authors have seen and agree with the contents of the manuscript and there is no financial interest to report.

AUTHOR'S CONTRIBUTIONS

Conceptualization, TAN. and TT.; methodology, PRA..; validation, CM., formal analysis, TAN.; investigation, TAN.; resources, TT.; writing—original draft preparation, TAN.; writing—review and editing, SM., ZSG, CM; visualization, SM., ZSG, CM.; supervision, CM., SM.; project administration, PRA.;. All authors have read and agreed to the published version of the manuscript.

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