- CASE SERIES —

The Prenatal Diagnosis and Perinatal Outcomes of Fetal Intra-Abdominal Cysts

Fetal İntra-Abdominal Kistlerin Prenatal Tanı ve Perinatal Sonuçları

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ABSTRACT

ÖZET

Objective: The aim of this study is to evaluate the perinatal ultrasonographic findings and perinatal outcome after prenatal diagnosis of fetal abdominal cysts.

Method: Fetuses diagnosed with abdominal cysts between 2008 and 2013 were retrospectively analysed. We analyzed maternal, fetal and perinatal variables for all cases.

Results: Eighteen fetuses 3 of that are males (16.7%) and 15 are females (83.3%) with the diagnosis of fetal abdominal cyst were analysed. The mean gestational age at diagnosis of a fetal abdominal cyst was 26.78 ± 6.75 weeks. The mean diameter of fetal abdominal cyst was 39.81 ± 20.47 mm at the time of diagnosis. There were 15 liveborn cases and two intrauterine deaths. One case was terminated due to multiple anomalies. Fetal abdominal cyst was resolved in four cases during antenatal follow-up period. Surgery was required in six cases neonatally (40%). Overall spontaneous mortality was 3/17(17.6%), of which two cases were intrauterine and one case was postoperative.

Conclusion: Parents should be informed about associated anomalies, perinatal, and postnatal outcomes of fetal intra-abdominal cysts. These results may be useful for prenatal counseling of fetal abdominal cyst.

Keywords: fetus; abdomen; cysts; ultrasonography; prenatal diagnosis; pregnancy outcomes

Amaç: Bu çalışmanın amacı, perinatal tanı alan fetal abdominal kist olgularının ultrason bulguları ve perinatal sonuçlarının değerlendirilmesidir.

Materyal ve Metod: 2008-2013 yılları arasında Fetal abdominal kist tanısı almış olgular retrospektif olarak incelenmiştir. Maternal, fetal ve perinatal veriler incelenmiştir.

Bulgular: 18 fetal abdominal kist olgusundan üçü (%16,7) erkek, 15'i (%83,3) kız fetustü. Fetal abdominal kist tanısı konulan ortalama gebelik haftası 26,78±6,75 hafta idi. Tanıda ortalama fetal abdominal kist çapı 39,81±20,47 mm olarak saptandı. 15 olgu canlı doğum tanısı ile, 2 olgu ise intrauterin ölüm ile sonuçlandı. Bir olgu multiple anomali nedeni ile termine edildi. Fetal abdominal kisti olan dört olgu intrauterin dönemde kayboldu. Altı olgunun (%40) cerrahi müdahale gereksinimi oldu. Total spontan mortalite 3/17 (%17,6) olarak, iki olguda intrauterin dönemde, bir olguda postoperatif dönemde meydana geldi.

Sonuç: Aileler fetal abdominal kistlerle ilişkili anomaliler ve peri-postnatal sonuçları hakkında bilgilendirilmelidir. Bu sonuçlar fetal abdominal kist olgularının danışmanlığında kullanılabilir.

Anahtar Kelimeler: fetüs; karın; kistler; ultrasonografi; prenatal tanı; gebelik sonuçları

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INTRODUCTION

Fetal abdominal cysts (FAC) can originate from renal, intestinal, mesenteric, ovarian, hepatic, and biliary systems (1). In the differential diagnosis, it is difficult to define the precise etiology of FAC prenatally. Although FAC have mostly been reported in the second trimester, ones with gastrointestinal or ovarian origin may not be identified till the third trimester (2). Furthermore, cases with FAC identified in the first trimester have been reported (3). FAC may have different characteristics in ultrasonography and different locations that makes exact diagnosis, origin and prognosis unclear. They can regress or grow progressively until being operated in the postnatal period. The prenatal and postnatal diagnosis may be discordant in some cases. When cyst has a nonspecific origin; prenatal counseling, forecasting of postnatal treatment and management may be complicated (1, 2). The aim of the present study was to establish the accuracy of ultrasonography in identifying the origin of the FAC and help to determine outcome antenally.

MATERIAL AND METHODS

This study was performed as a retrospective case series on patients with a prenatal diagnosis of the FAC attending in the second or third trimester to Maternal Fetal Medicine Department of Zeynep Kamil Maternity and Training Hospital from 2009-2013. All patients had a detailed morphology scan and Doppler flow studies as appropriate. All ultrasound examinations were performed on Voluson 730 Expert or Voluson 730 Pro (GE Healthcare, Milwaukee, WI). Gestational age was determined by the last menstrual period (LMP) and confirmed by the first or the second trimester ultrasound examination. Cysts arising from renal system and bowel dilatation were excluded. All cases were delivered in our clinic. The data was collected retropectively from written hospital records, the hospital database system, and via telephone interviews when needed. The characteristic appearance of fetal cyst was defined as a uniloculated simple cyst, biloculated and multiloculated-complex cyst. Uniloculated simple cyst was defined as a cyst without septa and solid component (Figure 1). Biloculated cyst was identified as a cyst with septa or adjacent two cysts (Figure 2). Multi-loculated complex cyst is described as a cyst with multiple septae containing hemorrhage or solid component (Figure 3). Also, size and location depending on the abdominal origin (right - left - midline / upperlower) were defined. Obstetric and perinatal outcomes were evaluated as gestational age at birth, mode of delivery, birth weight, postnatal

diagnosis, need for surgery, cyst resorption time and intrauterine fetal demise. Autopsy was performed on intrauterine fetal loss cases as well as the terminated ones. Statistical analysis was performed using SPSS, v11.5 (SPSS, Inc., Chicago, IL).



Figure 1. Uniloculated simple cyst without septa and solid component.



Figure 2. Biloculated cyst with septa and adjacent two cysts.





RESULTS

Eighteen fetuses, 3 males (16.7%) and 15 females (83.3%) fetal abdominal cyst with undetermined origin were recorded. All cases were diagnosed during the second and the third trimesters. The mean maternal age and gestational age at diagnosis of a fetal abdominal cyst was 26.17±3.62 years range [22-35] and 26.78±6.75 weeks [16-36], respectively. The mean diameter of fetal abdominal cyst was 39.81±20.47 mm [16.5-82.5] at diagnosis. There were 15 live born cases. The median gestational age of live born cases was 38.73±3.22 weeks [28-42] and the mean birth weight was 3056.0±585.6 grams [1200–3580]. There were two neonatal deaths. One of these fetuses had uniloculated cvst with bilateral hydronephrosis, bladder extrophy and anal atresia. The second fetus had multiloculated complex cyst with megavesica, ascites, and anhydramnios. In the terminated case at 18 weeks of gestation, fetal anomalies were pericardial effusion, Ventricular septal defect (VSD), flexion contracture of the hands and single umbilical artery.

Demographical characteristics, gender, cystic morphology, cystic location, prenatal diagnosis, postnatal diagnosis and outcome of cases were demostrated in Table 1. The cases were grouped as uniloculated, biloculated, and multi-loculated complex cyst according to their antenatal ultrasonographic appearance. Uniloculated cyst was noted in 10 (55.6 %) cases. The diagnosis was confirmed in eight cases postnatally. Four ovarian cysts, one hepatic cyst, one anal atresia, one meconium ileus, and one were enteric duplication cyst. Two cases one of which were antenatally considered as ovarian cyst and the other as mesenteric cyst resolved during intrauterine life. The most common localization of uniloculated cysts were in lower abdomen. Biloculated cyst was noted in 4 (22.22 %) cases. The diagnosis was confirmed in three of the cases postnatally. One of them with ovarian cyst and two were anal atresia. One case considered as ovarian cyst disappeared during antenatal period. All biloculated cysts were at lower abdomen. Multi-loculated complex cyst was noted in 4 (22.2%) cases. Postnatal diagnoses of three cases were ovarian cyst filling whole fetal abdomen in one; anal atresia in other; and megavesica, pancretic and biliary system cyst in the third case. One case which was considered as mesenteric or duplication cyst prenatally disappeared during antenatal period. One of the multi-loculated cysts was located on the upper left abdomen, two of them filled the whole abdomen and one was located on the lower right abdomen. The most common antenatal diagnosis was ovarian cyst. Ovarian cyst cases were

diagnosed at an average of 30.13±5.59 [20-36] gestational weeks. Diagnosis was confirmed as ovarian cyst at 6 of the 8 cases postnatally. Two cases were resolved during antenatal follow-up. Two of the six persistent ovarian cyst cases had operation, while four cases were regressed at postnatal period. Resolution time of this group was 2-9 months. Cases that required operation postnatally has an average cyst diameter of 65.25±0.35 mm. Cases with prenatal and postnatal resolution of ovarian cysts had an average of 17.0±0.71 mm and 33.63±4.42 mm, respectively. Pathologic evaluation revealed benign functional cyst in two cases who underwent cystectomy due to ovarian torsion in early neonatal period. The cysts were defined as "nonspecific fetal abdominal cyst" if the origin of the cyst is not certain. There were five cases in this group which consisted of the diagnoses of anal atresia in three cases, enteric dublication cyst in one case, and complex anomaly including urethral atresia, megavesica, pancreas and bilier cyst in one case after postnatal evaluation. One case had a postnatal diagnosis of meconium ileus although prenatal diagnosis was mesenteric cyst. This case was presented with a simple cyst about 51x23 mm in diameter at 22th gestational age. Ultrasound examination revealed no additional anomalies, but ascites was detected at 28th week even though the cystic mass regressed. The case had emergency cesarean section due to fetal distress. Colostomy was performed because of meconium peritonitis due to distal colon perforation. The sweat test was found negative for this case. Unfortunately the case died of sepsis when he was 3.5 months old.

11 of 18 fetuses had isolated cyst, while remaining seven fetuses have associated other anomalies including one cloacal extrophy with bilateral hidronephrosis, one multicystic dysplastic kidney, two bilateral hidronephrosis, one abdominal ascites, one megavesica and ascites, one pericardial effusion, VSD, flexion contracture of the hands and single umbilical artery. Four of the cases who had diagnosis of anal atresia also had additional anomalies. Three of them had genitourinary tract anomalies. Two cases with severe anomalies were detected in early gestational age (16 and 18 weeks), two cases who had only hydronephrosis were detected as late as 34 and 39 weeks. Karvotype analyses were performed in two cases with additional anomalies. No karyotype abnormality was detected. Overall spontaneous mortality was 3/17 (17.6%), of which two cases were intrauterine and one case was postoperative mortalities. Six cases (40%) of 15 live births were operated postnatally. These cases included two ovarian cysts, two anal atresia, one enteric duplication cyst and one meconium ileus.

	Outcome	Good (4 years old)	an	resolved in the 4th month (4 years old)	6th month resolved, nephrectomy (4 years old)	Good (3 years old)	Waiting for the second opera- tion (1 year old)	3.5th month sepsis, exitus	Unchanged follow-up (6 years old)	9th mounth resolve (3 years old)	Good (5 years old)	Good (2 years old)	۵N	TOP	Good (6 years old)	Good (4 years old)	Good (5 years old)	resolved in the 9th month (2 years old)	Good (5 years old)	eous Vaginal Velivery,
graphic and clinical data in 18 fetuses with fetal intra-abdominal cysts.	Surgery	Cystectomy at the 5th day, benign	Q	Q	Q	Inoculation in the 3rd month	Colostomy in the 2nd day	Colostomy in the 2nd day	No	No	Cystectomy in the 3rd day	Colostomy in the 1st day	No	Q	No	No	No	Q	N	ISD: Normal Spontane
	Postnatal diagnosis	Ovarian cyst	Cloacal extrophy+ Anal atresia	Ovarian cyst	Ovarian cyst	Enteric duplication cyst	Anal atresia	Meconium ileus	Isolated hepatic cyst	Ovarian cyst	Ovarian cyst	Anal atresia	Urethral atresia, megavesica, pancreatic cyst and bilier cyst with complex anomaly	Anal atresia	Disappeared	Disappeared	Disappeared	Ovarian cyst	Disappear	emise, TOP: Termination of Pregnancy, N
	Delivery, fetal outcome	CS at 40weeks, 3200 g	NSD at 30 weeks, 1300 g	CS at 40 weeks, 3450 g	NSD at 42 weeks, 3030 g	NSD at 41 weeks, 3350 g	NSD at 38 weeks, 3040 g	CS at 28 weeks, 1200 g	CS at 41 weeks, 3580 g	CS at 39 weeks, 3000 g	CS at 40 weeks, 3500 g, distosia	NSD at 39 weeks, 3200 g	IUD at 24 weeks, 610 g	TOP at 18 weeks, 210 g	Cs at 39 weeks, 3300 g	Cs at 39 weeks, 3300 g	CS at 37 weeks, 2540 g	CS at 39 weeks, 3400 g	CS at 39 weeks, 2750 g	I Cyst, IUD: Intrauterine De
	Antenatal diag- nosis	Ovarian cyst	Cloacal extrophy, nsFAC	Ovarian cyst	Ovarian cyst	nsFAC	nsFAC	Mesenteric cyst	Hepatic cyst	Ovarian cyst	Ovarian cyst	Mesenteric? Ovarian cyst?	nsFAC	nsFAC	Ovarian cyst	Mezenter cyst ? Duplication cyst?	Ovarian cyst	Ovarian cyst	Mesenteric?	fic Fetal Abdomina
	Additional findings	Q	Bilateral hydronephrosis, bladder extrophy	Polyhydramnios	Mutticystic dysplastic kidney	Ŷ	Bilateral Pelviectasis	Ascites	Q	Q	N	Bilateral hydronephrosis	Megavesica, ascites, anhyd- ramnios	Pericardial effusion, VSD, flexion contracture of hands, single umblical artery	N	9	Oligohydramnios	Q	Oligohydramnios	asonography, nsFAC: non-speci
	Cyst diameter (mm)	70×60	30×30	36x30	40x40	38x19	63x73	51x23	50x45	30x33	60x61	65x70	80x85	24x12	17x16	20x17	20×17	30x30	25x23	USG: Ultra
	USG Localization	Left lower	Middle lower	Right lower	Left lower	Left upper, between stomach and spleen	Left lower	Midline, Iower	Right upper	Left lower	Midline, fills abdo- men	Right lower	Midline, fills abdo- men	Left lower	Middle lower	Left upper	Right lower	Right lower	Right upper	M: Male, F: Female,
	NSG	Uniloculated	Uniloculated	Uniloculated	Biloculeted	Uniloculated	Biloculeted	Uniloculated	Uniloculated	Uniloculated	Complex	Complex	Multiloculated complex	Biloculeted	Biloculeted	Multiloculated	Uniloculated	Uniloculated	Uniloculated	P: Parity, Sex:
sonoc	GA	35	8	36	58	53	8	8	25	20	31	34	23	9	26	17	29	36	30	sravida,
ary of	Sex	ш	ш	ш	ш	ш	ш	Σ	ш	ш	ш	ш	ш	Σ	ш	ш	ш	ш	Σ	e, G : G
1. Summ	MA, GP	26, G2P1	29, G2P1	29, G1P0	23, G1P0	22, G5P1 A4	23, G1P0	22, G1P0	35, G2p1	26, G1p0	28, G2P1	23, G1P0	26, G1p0	30, G2P0	28, G1P0	22, G1P0	23, G1P0	30, G1P0	26, G1P0	Aaternal Age
Table	Case	-	N	e	4	5	9	2	œ	o	10	7	12	13	4	15	16	17	18	CS: C

DISCUSSION

The detection of FAC is quite easy with ultrasonography; however, it is still hard to detect its origin. Even though there is no distinct algorithm to identify the origin of FAC; location of the cyst and its ultrasound appearance can help to reveal its origin. FAC is often detected in the second and the third trimester. Differential diagnosis should include gastrointestinal tract (mesenteric, omentum, intestinal duplication, hepatic, biliary cysts and meconium pseudo-cysts) and genitourinary tract (ovarian, renal, urachal and adrenal cyst) (4). Additionally, extra-abdominal pulmonary, spinal or retroperitoneal lesions can resemble FAC (5, 6).

The ovarian cyst is the most common FAC lesion detected in female fetus. Similarly, in this study ovarian cyst was found the most frequent diagnosis of the FAC lesions in female fetuses (53.3%) at the antenatal period. Slodki et al. detected ovarian cyst ratio as 74% in FAC cases (7). We found general ovarian cyst ratio in all FAC lesions as 44.4%. D'Addrio et al. studied ultrasonography findings of fetal ovarian cysts in 25 pregnant women. The most frequent antenatal finding was uniloculated simple cyst at lower lateral abdomen during the third trimester of pregnancy. Also, they emphasized that it could be rarely in a complex composition secondary to torsion and hemorrhage (8). Similarly, 62% of our ovarian cyst cases were uniloculated and 87.5% was located in the lower abdomen. We reported that two cases had complex cystic structure because of hemorrhage confirmed via operation in the neonatal period. Prognosis of ovarian cysts is associated with cyst size and structure. Bagelon et al. have studied 73 ovarian cyst cases prospectively and reported that 75% of cysts less than 50 mm size resolved in utero (9).

Slodki et al. has identified that spontaneous regression rate of the <50 mm cyst was 98% and complication rate of >50 mm kist was 93% on 420 fetal ovarian cyst cases diagnosed antenatally. 25% of our cases resolved in utero while 50% resolved in the postnatal period. Thus intrauterine and postnatal resolution is not rare in ovarian cysts and should be considered in prenatal counselling. In addition, our findings are also consistent with the current literature regarding to postnatal surgery requirement (10). Our study reported that the cyst >50 mm in the postnatal period necessitated surgery, cysts <50 mm had spontaneous regression. Even in utero, spontaneous regression is observed in cysts <20 mm. Schenkman et al. reported that ovarian cysts with diameter of <40-50 mm resolved spontaneously without need for surgery within the first 4-5 mounts of postnatal life (11).

Rarely, ovarian cysts enormously expand causing fetal abdominal enlargement and may cause dystocia (8). In this study, one case with the cyst filling the whole abdomen had an abdominal circumference >95th centile and applied cesarean section for the possibility of dystocia. The rates of chromosomal and non-chromosomal anomalies in ovarian cyst cases are very low. Shodki et al. reported 87% of 27 patients with ovarian cysts were isolated. In our study 87.5% of diagnosed ovarian cyst cases were isolated in accordance with the literature and no chromosomal anomaly was detected in the postnatal period (7).

Another important issue about FACs is the association with gastrointestinal tract anomalies such as imperforated anus which can be presented with FAC. Brandberg et al. reported that only 11(15.9%) of 69 cases with imperforated anus were diagnosed prenatally. In all 11 fetuses, a dilatation of the rectum or lower part of the bowel was present and two had additional intraluminal calcifications. Additional anomalies including mostly urogenital and cloacal were present in 85.5% of the cases. The karyotype was abnormal in 9 (13%) of the cases (12). Ochoa JC at el. analyzed 189 cases with high risk of anal atresia in second and third trimester. They detected 14 cases of anorectal malformation, 9 (64.3%) had prenatal dilatation of the distal bowel and 5 (35.7%) had intraluminal calcified meconium or enterolithiasis. Also, they reported that absent fetal perianal muscular complex on prenatal sonography in this high-risk population had a sensitivity of 100%, specificity of 99%, true-positive rate of 93%, and false-positive rate of 7% for the diagnosis of anorectal atresia (13). In our study no definitive diagnosis of anorectal atresia was possible in antenatal period. Three out of four anal atresia cases had nonspecific FAC lesions antenally. One case was misdiagnosed as mesenteric or ovarian cyst but the postnatal diagnosis came out to be anal atresia. Although morphologies of all cystic masses were different, all appeared in the lower abdomen during antenatal ultrasound scanning. Additional anomalies were found in four cases, in accordance with the literature 75% were originating from the urogenital system. None of these four cases had polyhydramnios. Although anorectal malformations were reported to be common in males (14), our study revealed three out of four cases to be female. For this reason when FAC is identified at the lower abdomen with additional anomalies in female fetuses, lower gastrointestinal tract obstruction should be kept in mind.

Another rare etiology of FAC is enteric duplication cysts, antenatal diagnosis of which are challenging because of nonspecific structure and localization (15, 16).

In this study, one case with a simple cyst in the upper abdomen localized between stomach and spleen was followed-up as nonspecific FAC, admitted to the hospital with vomiting and abdominal distention in the early postnatal period, operated and diagnosed to have gastric duplication cyst. Richard et al. recommended ultrasonographic criteria that may help for specific diagnosis of two enteric duplication cysts. They declared that presence of thick muscular wall, being solitary and observed peristalsis may help to distinguish these cysts from ovarian, mesenteric, omental and other abdominal cysts (15). Also Meyberg et al. reported a case with solitary cyst on right lower abdomen and needle aspiration was applied twice in the antenatal period. The case admitted to the hospital with vomiting and abdominal distention in the postnatal period was operated with the diagnosis of duplication cyst of the colon (16). Eventually, prenatal diagnosis is important for appropriate management before appearance of symptoms or complications.

Differential diagnosis of upper abdominal cystic masses should include choledochal or hepatic cysts besides proximal gastrointestinal tract obstruction. Hepatic cysts are rare and few cases have been described in the literature. Macken et al. reported a case considered to be hepatic cyst presenting with right upper quadrant cystic masses anterior to the gallbladder, demonstrating no peristalsis or blood flow. The cyst was inseparable from the liver and caused some flattening of the liver edge. Postnatal sonographic examination confirmed congenital hepatic cysts (17). Additionally, Bronsen et al. pointed out that disappearance of hepatic cyst can be expected especially for small and peripheral hepatic cysts (18). In our study one simple cyst in the right upper abdomen without blood flow was considered as hepatic cyst antenally and this diagnosis was confirmed postnatally. Meconium peritonitis is extremely rare and only small series and case reports were reported. Chan et al. reported seven meconium peritonitis cases. 43% of their cases presented with ascites and calcification, or dilated/hyperechoic bowel loops. The other 43% had only ascites in ultrasound examination. Two patients' CT scans showed persistent intestinal perforation not visible with prenatal USG, and required urgent surgery (19). Saleh et al. declared that the prenatal diagnosis of meconium peritonitis was confirmed by clinical and radiological findings in 8 (62%) of 13 live born infants (20). Our case was considered as simple mesenteric cyst in the mid pelvis on the 22nd gestational weeks, but only ascites was apparent on the 28th gestational weeks. In postnatal period, it was postoperatively detected to be meconium peritonitis due to distal colon perforation. Although antenatal diagnosis of meconium cyst is hard, it should be kept in mind in such cases presenting with ascites, calcifications, and the changing nature of the cyst. Precise diagnosis of meconium cyst is very important due to emergent surgery requirement.

In our series, surgery was required in 40% of live births. Similarly, Özyüncü et al. and Sherwood et al. reported surgery rate in FAC as 34.4% and 29%, respectively (1,21). We reported that overall spontaneous mortality rate was 3/17 (17.6%). Accordingly, Özyüncü et al. reported overall spontaneous mortality rate to be 11/64 (17%). Intrauterine deaths in our series were 2/17 (11.8%), while this was found to be 3.6% and 4.2% in previous reports, respectively (1, 21). We think this difference arises from the inclusion criteria of the study groups. Sherwood et al. included only intrauterine losses while Özyüncü et al. included upper gastrointestinal and urinary system anomalies.

As a result, ultrasonography may not be sufficient to detect the origin of FAC lesions. But the appearance and localization of the cvst. additional fetal anomalies, fetal gender, presence of peristalsis in the cyst, and presence of thick muscular wall are important clues for the diagnosis and management of the cyst. Furthermore, detection time of the cyst as gestational age and the changes in the follow-up will be helpful in the surveillance. Another important issue to be mentioned in prenatal counselling is the diagnostic difficulty of the origin of FAC especially when additional anomalies are present. Families should be informed of the probable association with lower gastrointestinal tract anomalies.

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