Splenic cyst as a rare cause of fetal abdominal cystic mass: A multicenter series of nine cases and review of the literature

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Abstract

Introduction: Congenital splenic cysts are a rare cause of abdominal cystic masses in the fetus. We describe nine cases of this condition, the largest reported experience to date. A review of the literature for similar cases was also performed.

Methods: Cases of congenital splenic cyst were collected from three prenatal diagnostic centers and from a dedicated website of prenatal diagnosis. Information regarding clinical and sonographic findings was obtained by reviewing ultrasound reports and medical records. An electronic search of the Pubmed/MEDLINE database for similar cases, with subsequent manual cross-referencing, was performed and the relevant information from the articles was retrieved.

Results: A total of nine cases were added to the currently existing literature of 32 cases. In all but one of our cases, the prenatal detection of the splenic cyst was made in the third trimester, with a median gestational age at diagnosis of 30 weeks (range 22–37). The splenic cyst was confirmed after delivery in all but one case, which was lost to follow up. Postnatal sonographic examinations performed in the remaining eight cases showed that the cyst increased in size in two (25%), was stable in size in one (12%), decreased in size in one (12%), and completely regressed in the other four cases (50%). No complications associated with the cysts were reported in the infants.

Conclusions: Congenital splenic cyst appears to be a benign condition with no known risk of perinatal complications. Nevertheless, differential diagnosis with other fetal cystic masses of the left upper abdominal quadrant and postnatal sonographic follow up to monitor the size of the cyst are important issues to be considered in the perinatal management of these cases.

Keywords

Abdominal cyst, fetal ultrasound, prenatal diagnosis, splenic cyst

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Introduction

Prenatal sonographic detection of an abdominal cystic mass poses clinically significant diagnostic and management challenges.^{1–3} Fetal abdominal cysts are mainly related to anomalies of the gastrointestinal or urogenital tracts and their differential diagnosis and prognosis depend on their sonographic characteristics, location,

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and subsequent antenatal course.¹⁻⁴ Congenital splenic cysts are among the less frequent causes of abdominal cystic masses in the fetus. In this report, we describe a series of nine cases diagnosed prenatally by ultrasound and discuss the presenting sonographic features, antenatal and neonatal management, perinatal implications, and outcome. A review of the literature for similar cases is also presented.

Material and methods

This was a retrospective review of cases of congenital splenic cyst detected prenatally by ultrasound. Cases were collected by reviewing the ultrasound databases from three Fetal Medicine centers in Chile and Argentina over a 15-year study period from 2002 to 2016. In addition, cases summarized on an electronic website for prenatal diagnosis (www.thefetus.net) were also identified and the authors contacted and invited to collaborate with this series.

Prenatal diagnosis of a congenital splenic cyst was based on the sonographic detection of a round or oval anechoic structure with smooth walls located in the left upper abdominal quadrant posterior to the stomach (Figure 1). In addition, color Doppler ultrasound was used to rule out the presence of blood flow signals within the lesion (Figure 2). Although in some cases it was difficult to demonstrate that the cyst was located within the spleen at the initial examination, thorough examination upon referral revealed that the other abdominal organs including the stomach, liver, left adrenal gland, and left kidney appeared normal and were clearly separated from the cystic mass, making the diagnosis of splenic cyst straightforward. Information on maternal demographics, sonographic features, subsequent antenatal management, and

perinatal outcome was obtained by reviewing the ultrasound reports and images, medical records, and, when required, by contacting the referring obstetrician or the patient herself as part of our standard clinical care. Due to the retrospective nature of this research, permission of the Institutional Review Board was waived.

An extensive review of the literature was also performed through a computerized search of the Pubmed/MEDLINE database combining the terms “splenic cyst” and “spleen cyst” with “fetal”, “fetus”, “prenatal diagnosis”, and “antenatal diagnosis” without limits of time and language. Search results were also cross-referenced to identify additional articles.

Results

A total of nine cases of congenital splenic cysts were identified, including three that have been partially reported upon previously. Table 1 presents the most relevant clinical and sonographic findings in these cases. The median maternal age was 32 years (range 22–39) and four (44%) of the women were primigravids, four (44%) were parous, and one (11%) was of unknown parity. The median gestational age at diagnosis was 30 weeks (range 22–37). Only one (11%) case was detected during the routine second-trimester ultrasound examination at 22 weeks; the remaining eight (89%) were detected during the third trimester. In all cases detected in the third trimester, the second-trimester anatomy scan had been reported as normal. Indication for the third-trimester scan included a variety of reasons unrelated to the presence of the cyst, mainly surveillance of fetal growth and wellbeing and assessment of amniotic fluid volume.

Figure 1. Transverse sonographic view of the fetal left upper abdomen at 35 weeks shows a single cystic mass (c) measuring 22 mm within the spleen (Sp).

Figure 2. Color-Doppler ultrasound at 28 weeks in a fetus with a 7-mm splenic cyst (between calipers) shows no blood flow signals within the mass. Sp: spleen; St: stomach.

Table 1. Prenatal detection of fetal splenic cyst: Clinical cases

Case	MA (years)	Parity	GA at diagnosis (weeks)	Cyst			Associated anomalies	Outcome
				Type	Size (mm)	Prenatal course		
1 ^a	29	0	22	Single	11	NA	No	Term delivery, female, healthy. Cyst of similar size at 1 month
2 ^b	22	1	28	Single	8	10 mm at 37 weeks	No	Term delivery, female, healthy. Cyst resolved at 2.5 years
3	32	1	28	Single	7	NA	No	Term delivery, healthy. Cyst resolved postnatally
4	29	1	30	Double	12 and 10	13 and 12 mm at 32 weeks and 16 and 14 mm at 36 weeks	No	Term delivery, male, healthy. Cysts increased to 22 and 15 mm at 2 days and to 22 and 17 mm at 4 months
5	31	0	30	Single, septated	17	NA	No	Term delivery, healthy. Cyst resolved at 1 month
6 ^c	34	2	34	Single	11	NA	No	Term delivery, male, healthy. Cyst of similar size at 3 months. Resolution at 2 years
7	33	0	35	Single	14	NA	Left megaureter	Term delivery, male, left megaureter. Cyst of similar size postnatally and decreased to 8 mm at 9 months
8	39	NA	35	Single	22	NA	No	Term delivery, male, healthy. No follow-up information available
9	36	0	37	Single	8	NA	No	Term delivery, healthy. Cyst increased to 11 mm at 2 months

MA: maternal age; GA: gestational age; NA: information not available.

^a<https://www.sonoworld.com/TheFetus/page.aspx?id=3649>^b<https://www.sonoworld.com/TheFetus/page.aspx?id=2092>^c<https://www.sonoworld.com/TheFetus/page.aspx?id=2460>

In eight of the nine cases, only a single cyst was present, which measured between 7 and 22 mm in maximum diameter at the time of presentation. Only one of these cases had a prenatal follow-up scan, at which time the cyst was seen to minimally increase in size from 8 mm to 10 mm. In the remaining case, two distinct cystic masses measuring 12 mm and 10 mm in diameter were visualized close to each other within the fetal spleen. Both cysts were noted to increase in size on subsequent scans and at the neonatal abdominal scan (Figure 3). In one fetus with a single cyst, a septation within the cyst was identified (Figure 4); in all the other cases the content was homogeneously anechoic. Associated anomalies were noted in only one case in which an ipsilateral megaureter was identified; the

renal anomaly was considered as an incidental finding and not related to the splenic cyst. All fetuses were delivered at term; however, information on the type of delivery was available only in eight cases and all but one woman delivered vaginally. In the case delivered by Cesarean section, the indication was unrelated to the presence of the cyst. The physical examinations and clinical courses of the neonates were normal in all cases. Neonatal abdominal ultrasound and subsequent pediatric sonographic follow up was available in all but one case. The splenic cyst was confirmed in all these eight cases and subsequent sonographic examinations showed that the cyst increased in size in two (25%), was similar size in one (12%), decreased in size in one (12%), and completely regressed in the remaining four cases (50%). No complications associated to the cysts were reported in the infants.

Review of the literature produced 32 additional cases,⁵⁻²⁵ of which 22 were reported in English,^{5,8-11,13,15-17,20-24} six in Spanish,^{7,18,19,25} three in French,^{6,14} and one in Italian.¹² Table 2 summarizes the clinical and sonographic information obtained from the review of these cases.

Discussion

The prenatal diagnosis of a congenital splenic cyst was first reported by Lichman and Miller⁵ in 1988. In their report, a 13 mm cyst was detected in the left upper abdominal quadrant at 33 weeks of gestation. Although the splenic cyst was confirmed postnatally by ultrasound and no changes in size or shape were noted, information on long-term follow up was not available. A case with a large splenic cyst measuring 45 mm in diameter detected at 32 weeks was reported the following year.⁶ As the mass was seen to be distinct from the left kidney, the diagnoses of mesenteric cyst and extraparenchymal renal cyst were considered. A laparotomy with partial splenectomy was performed postnatally due to the increasing size of the cyst to 70 mm and displacement of the neighboring organs. Histopathological examination of the surgical specimen revealed an epidermoid splenic cyst.⁶

Subsequent reports, however, have demonstrated that the vast majority of fetal splenic cysts are relatively small and have a benign course in the fetus, remaining stable, decreasing in size, or even completely regressing at subsequent postnatal sonographic examinations. Indeed, Garel and Hassan¹⁰ reported that all three of their prenatally detected cases resulted in resolution of the cyst, an observation that has been subsequently repeated by several other authors (Table 2). Of note, all but one⁶ of the infants described in the antenatal literature have remained clinically asymptomatic without any complications attributable to the cyst.

Figure 3. Left parasagittal view of the fetal abdomen and thorax shows two fetal splenic cysts (c) measuring 16 mm and 14 mm at 36 weeks. The cysts were first detected at 30 weeks, at which time they measured 12 mm and 10 mm, respectively.

H: heart; Sp: spleen; St: stomach.

Figure 4. Fetal single splenic cyst (between calipers) measuring 17 × 15 mm at 30 weeks, containing a single septation (arrow).

Sp: spleen.

Table 2. Prenatal detection of congenital splenic cyst: Review of the literature

Authors, year	MA (years)	Parity	GA at diagnosis (weeks)	Cyst		Size (mm)	Prenatal course	Associated anomalies	Perinatal outcome and remarks
				Type					
Lichman and Miller, 1988	34	2	33	Single		13	Similar size at 38 weeks	No	Term delivery. Cyst confirmed after birth. No follow-up information available
Todde et al., 1989	NS	1	32	Single		45	61 mm at 37 weeks. Mesenteric cyst? Renal cyst?	No	Term delivery, firm mass in the left hypochondrium and epigastric region. Cyst of 70 mm at postnatal scan and CAT. Partial splenectomy on day 3. Pathology: Epidermoid cyst. Splenic cyst not considered in the differential diagnosis
Badia Barnusel et al., 1990	23	Gravida 2	3rd trimester	Single		21	NS	No	Term delivery, male, healthy. Cyst decreased to 15 mm at postnatal scan. Resolution at 5 months
Stiller et al., 1991	29	0	35	Single		15	NS	No	Dichorionic-diamniotic twin. Cyst of similar size at 3 month
Tsao and Jeanty, 1993	30	0	33	Single		5	NS	Cleft lip and palate ^a	Term delivery, male, SGA, cleft lip and palate. No confirmation after delivery. Asymptomatic infant at long-term follow up
Garel and Hassan, 1995	NS	NS	32	Single		10	13 mm at 37 weeks	No	Cyst increased to 20 mm at day 10, and then decreased to 11 mm at 3 months. Resolution at 2 years
Garel and Hassan, 1995	NS	NS	28	Single		15	NS	No	Cyst of similar size (16 mm) at 1 month and 3 years
Garel and Hassan, 1995	NS	NS	33	Single		16	NS	No	Cyst of 18 mm at day 9, 17 mm at 3 months, and 17 mm at 6 months
Okada et al., 1995	32	1	17	Single		13	13 mm at 19 weeks, 7 mm at 21 weeks, 5 mm at 25 weeks	No	Term delivery, healthy. Cyst of 8 mm postnatally

(continued)

Table 2. Continued

Authors, year	MA (years)	Parity	GA at diagnosis (weeks)	Cyst			Associated anomalies	Perinatal outcome and remarks
				Type	Size (mm)	Prenatal course		
Bagnulo et al., 1996	30	0	36	Single	9	Similar size at 38 weeks	Talipes ^a	Term delivery, female, SGA, left talipes. Cyst of similar size at 1 month
Yilmazer and Erden, 1998	26	0	31	Single	12	Similar size at 35 weeks and 14 mm at 38 weeks	No	Term delivery, female, healthy. Cyst increased to 17 mm at postnatal scan and at 2 months and decreased to 13 mm at 4 months. Resolution at 7 months
Kabra and Bowen, 2001	30	0	20	Single	6	Similar size at 29, 35, and 38 weeks	No	Term delivery, male, SGA, otherwise healthy. Cyst of 6 mm postnatally and of 4 mm at 5 and 10 months
Lopes et al., 2001	24	0	25	Single	11	14 mm at 28 weeks, 22 mm at 31 weeks, 23 mm at 34 weeks, and 31 mm at 38 weeks	No	Term delivery, female, healthy. Cyst of 30 mm postnatally, 25 mm at 2 months, and 5 mm at 5 months. Resolution at 6 months
Brottier et al., 2001	25	1	32	Single	14	19 mm at 34 and 22 mm at 36 weeks	No	Term delivery, healthy. Cyst of 28 mm postnatally. Resolution at 6 months
Brottier et al., 2001	25	0	32	Single	Few mm	Few mm at 34 and 36 weeks	No	Term delivery, healthy. Cyst of 30 mm at day 24, 35 mm at day 31, and 34 mm at 9 months and 2 years
Saada et al., 2006	26	0	23	Single	10	15 mm at 33 weeks	No	Term delivery, female, healthy. Cyst of 11 mm postnatally. Resolution at 2 months
Saada et al., 2006	33	0	25	Single	10	Similar size at 30 weeks	No	Term delivery, male, healthy. Cyst confirmed postnatally. Resolution at 3 months

(continued)

Table 2. Continued

Authors, year	MA (years)	Parity	GA at diagnosis (weeks)	Cyst			Associated anomalies	Perinatal outcome and remarks
				Type	Size (mm)	Prenatal course		
Catarina Prior et al., 2006	NS	1	34	Double	23 and 18	NS	No	Term delivery, male, healthy. Cysts of 24 and 19 mm at postnatal scan and of 27 and 23 mm on day 8 by CAT. Resolution at 5 months
Catarina Prior et al., 2006	NS	1	30	Single	15	NS	No	Term delivery, female, healthy. Cyst of 12 mm postnatally. Complete resolution at 2 years
Gomez-Suarez et al., 2006	28	NS	20	Single	10	17 mm at 22 weeks, 18 mm at 23 weeks, 3 mm at 35 weeks	No	Term delivery, male, healthy. Two cysts of 8 mm each at postnatal scan. Resolution at 5 months by MRI
Chen et al., 2007	37	2	26	Single	17	12 mm (15 mm by MRI) at 30 weeks and 14 mm at 34 weeks	No	Term delivery, female, healthy. Cyst of 8 mm by postnatal scan and 7 mm by postnatal CAT. Resolution at 5 weeks
Cuillier et al., 2007	29	2	27	Single	10 ^b	MRI at 30 weeks. Size NS	No	Term delivery, healthy. Cyst of 5 mm postnatally. Complete resolution at 3 months
Cuillier et al., 2007	25	NS	32	Single	12 ^b	12 mm at 37 weeks	No	Term delivery. Cyst confirmed postnatally. Complete resolution at 2 months
Cuillier et al., 2007	30	NS	30	Single	12 ^b	Confirmed at 36 weeks	No	Term delivery, healthy. Cyst confirmed postnatally. Resolution at 3 months
Cuillier et al., 2007	24	NS	33	Single	15 ^b	NS	No	Term delivery, healthy. Cyst confirmed postnatally. Complete resolution at 3 months
Dankovcik et al., 2009	30	3	27	Single	25	Similar size at 31 weeks	No	Term delivery, healthy. Cyst of similar size postnatally. Decreased to 10 mm at 3 months. Resolution at 6 months

(continued)

Table 2. Continued

Authors, year	MA (years)	Parity	GA at diagnosis (weeks)	Cyst			Associated anomalies	Perinatal outcome and remarks
				Type	Size (mm)	Prenatal course		
Dankovcik et al., 2009	39	0	31	Single	9	Similar size at 38 weeks	No	Term delivery. Cyst confirmed postnatally. Resolution at 6 months
Dankovcik et al., 2009	25	0	34	Single	20	NS	No	Resolution at 6 months
Venkataraman, 2009	29	NS	23	Single	12	14 mm at 29 weeks and 35 weeks	No	Term delivery, female, healthy. Cyst of 12 mm at 3 weeks and 7 mm at 5 months. Resolution at 13 months
Having and Anderson, 2010	Late 20s	0	36	Single	17	NS	No	Term delivery, female. No follow-up information available
Ruiz-Labarta et al., 2013	37	2	29	Single	7	Similar size through subsequent scans	No	Term delivery, male, healthy. Resolution at 9 months
Ruiz-Labarta et al., 2013	25	0	32	Single	8	Similar size through subsequent scans	No	Term delivery, female, healthy. Resolution at 3 months

MA: maternal age; GA: gestational age; NS: not stated; MRI: magnetic resonance imaging; CAT: computerized axial tomography; SGA: small for gestational age.

^aDiagnosis of associated anomaly missed prenatally.

^bInformation obtained from the corresponding author.

From the histopathologic point of view, splenic cysts are classified into two main categories, true cysts and pseudocysts, depending on the presence or absence of an epithelial lining, respectively. Pseudocysts are secondary to trauma or infection and occur mainly in adults.²⁶ True cysts, in contrast, are seen more frequently in children and are subclassified into parasitic or nonparasitic forms.²⁷ Congenital splenic cysts are categorized as a nonparasitic subtype, which includes epidermoid, dermoid, polycystic, or mesothelial (simple) cysts.²⁷ The pathophysiology of congenital splenic cysts is believed to be due to the involution of pluripotent cells in the splenic parenchyma with subsequent squamous metaplasia.⁹ However, confirmation of this theory is difficult to prove since all but one⁶ of the reported prenatal cases lack pathological examination of the splenic lesion. The reason for spontaneous resolution noted in some of the prenatally diagnosed cases remains unknown.

Congenital splenic cysts are rare although no accurate figures on their true incidence are available in the literature. This is supported by our review of the literature in which only 32 cases detected prenatally have been reported.^{5–25} However, as this has been mainly a third-trimester finding, and third-trimester ultrasound examination is not typically performed in the low-risk pregnant population, many cases of fetal splenic cyst likely remain undetected. Until now, the largest prenatal series was reported by Cuillier et al.²¹ who described four cases, followed by Garel and Hassan¹⁰ and by Dankovcik et al.,²² describing three cases each. Our series reports an even larger experience, although it was based on the recollection of cases from three referral centers for prenatal diagnosis in addition to those obtained from a dedicated website for prenatal diagnosis. Fetal splenic cysts generally manifest on ultrasound when the axial view of the fetal abdomen is obtained for abdominal circumference measurements. Our earliest diagnosis of splenic cyst was indeed diagnosed in that manner at 22 weeks. In cases in which there was doubt about the exact location, the diagnosis of a splenic cyst was confirmed at referral. Technical factors such as quality of the ultrasound machine, fetal position, maternal habitus, and experience of the operator at the initial scan as well as the rarity of this condition may account for the inability to make the exact diagnosis at initial presentation.

Fetal splenic cyst is typically an isolated finding; however, in one of our cases there was a left megaureter and in other reported cases a cleft lip and palate⁹ and talipes¹² were diagnosed postnatally, for an overall 7% (3 of 41) rate of associated anomalies. Although these anomalies seems to be unrelated to the splenic cyst, it is nevertheless recommended that a detailed fetal anatomic survey should be performed whenever this

anomaly is found to examine other organs to rule out other anomalies including systemic microcystic disease. After the prenatal identification of a suspected congenital splenic cyst, it is important that a differential diagnosis is formed because the prenatal diagnosis of other masses of the left upper abdominal quadrant may have different managements and prognoses.⁴ Differential diagnoses include cystic anomalies involving the upper abdomen such as duodenal obstruction, suprarenal cystic mass, intraabdominal cystic pulmonary sequestration, and polycystic kidney.⁴ Therefore, every effort to identify the precise location of the cyst should be performed. Although the use of fetal magnetic resonance imaging has been reported in two cases,^{20,21} it seems that the current resolution of modern ultrasound equipment is sufficiently accurate to determine the exact location, size, and appearance of the cystic mass.

Based on the available literature and our own experience, there is consensus that congenital splenic cyst is not anticipated to be associated with perinatal complications, although sporadic cases of rupture and hemorrhage have been reported in children and adults.^{10,27} Once detected prenatally, the parents should be counseled about the typically benign nature of this congenital anomaly, although prenatal follow up should be performed to confirm its exact location and monitor the size of the cyst. Postpartum ultrasound and pediatric follow up should be considered in order to rule out significant growth of the cyst in the postnatal period.

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Contributors

WS wrote the first draft of the manuscript and reviewed the literature. WS and AEW analyzed patient data and wrote the final version. WS, JHO, DC, FB, EA, and EYA contributed with case reports. All authors reviewed and approved the final version of the manuscript.

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