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Fetal Non-Ovarian Abdominopelvic Cystic Lesions: A Single-Center Report

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Fetal cystic abdominal lesions · Prenatal diagnosis · Cystic lesion resolution · Fetal cysts

Abstract
Introduction: There is a paucity of reports describing the clinical course and likely postnatal outcomes of prenatally identified simple cystic abdominopelvic lesions which are not associated with the ovary. ***Objective:*** The aim of this study was to describe the natural history and postnatal outcomes of prenatally discovered abdominopelvic cystic lesions seen at our center. ***Methods:*** This study is a retrospective review of all newborns with prenatally discovered non-ovarian simple cystic abdominal or pelvic lesions (September 2012–December 2018). Prenatal solid organ involvement, lesion size, and postnatal clinical outcomes are described. ***Results:*** Sixty-six patients with 68 cystic lesions were identified; 22 patients with 24 lesions met the defined study criteria and were included. Eleven (46%) resolved prenatally, while 5 (21%) resolved by 18 months of age. Of the 10 lesions associated with an organ, 4 (40%) resolved prenatally. Of the remaining 14 lesions not associated with a solid organ, 7 (50%) resolved prenatally. Seven lesions (29%) required postnatal surgical intervention. Larger maximum prenatal lesions tended toward postnatal surgical intervention (one-way ANOVA: $p = 0.072$). ***Conclusions:*** The majority of simple non-ovarian cystic abdominopelvic lesions at our center resolved in the perinatal period. Due to the low frequency of these lesions at fetal centers, a larger multicenter study based on a consistent monitoring protocol should be undertaken to better describe the resolution patterns of simple non-ovarian cystic lesions for improved prenatal counseling.

Introduction

Abnormal fetal lesions in the abdomen or pelvis may be discovered at the routine second trimester morphology screening or during the third trimester scans [1, 2]. Prenatal sonography of such lesions most often reveals a round anechoic or echogenic structure of varying size and position, and benign or malignant pathophysiologies cannot be differentiated. The prenatal management course of these patients is dictated by lesion location, involvement with anatomical structures, observed growth, and immediate risks to the fetus. Unfortunately, we currently have limited ability to anticipate whether the lesion will spontaneously resolve in the antenatal period, persist as a normal structural variant, or require surgical intervention after birth [3–5].

Fetal abdominal cystic lesions are relatively common, occurring in approximately 1 in 1,000 fetuses [3, 5–7]. Ovarian fetal cysts are one of the most common cyst types, and a large number of studies exist in the literature

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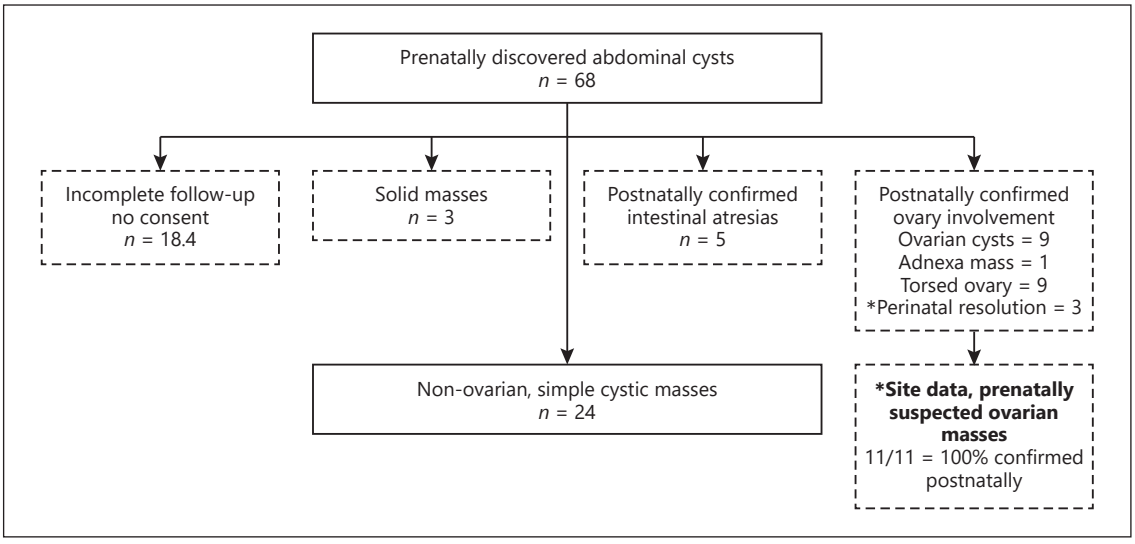

Fig. 1. Study population.

describing the unique properties and clinical course of ovarian cysts [2, 5]. Prenatally discovered fetal simple cystic lesions that are non-ovarian in origin are rarer and not as rigorously described in the literature. It has been reported that many such cysts spontaneously resolve and do not typically increase fetal morbidity or mortality, but the association between the initial prenatal diagnoses and the postnatal outcomes has been not thoroughly described [2, 5–8]. Data describing the natural history and postnatal clinical outcomes of such cysts could assist with prenatal counseling and outcome expectations for patients with this diagnosis. Therefore, this study reports our institutional natural history and outcomes for fetuses diagnosed prenatally with non-ovarian simple cystic abdominal or pelvic lesions. Prenatal sonographic lesion characteristics such as lesion size, solid organ involvement, and clinical outcomes are described for our center.

Materials and Methods

This study was conducted at Children's Minnesota and was approved by the Institutional Review Board (IRB). Cases of abdominal or pelvic cystic lesions discovered prenatally by ultrasound (US) between September 2012 and December 2018 were retrospectively reviewed. A trained staff member completed data extraction from the institution's electronic medical record, and extracted data were audited by a senior author (BAF).

The inclusion criteria included patients with findings of a fetal abdominal or pelvic cyst with at least one prenatal sonographic mass measuring >5 mm. After the initial detection, follow-up US evaluations were collected monthly or as often as clinically indicated to track lesion growth or regression; all available scan results for each lesion were reviewed for this study. In cases where we were able to identify the organ of involvement but could not give a definitive diagnosis based on imaging alone, we used the terminology of nonspecific diagnosis. For all lesions, even lesions that were no longer demonstrable by prenatal US, postnatal US was collected immediately after birth. If the lesion was postnatally identified and persistent, follow-up imaging was ordered as clinically indicated within 6 months to determine lesion growth patterns and whether a surgical strategy was necessary.

In this study, cystic lesions that were prenatally classified as solid or suspected to be ovarian or intestinal were excluded. Subjects without postnatal follow-up data related to the lesion were also excluded. Information related to solid organ involvement, lesion sizes, and postnatal clinical outcomes were collected. Study results were reported as summary statistics, and statistics testing for categorical or numerical data such as χ^2 analyses or ANOVA tests was applied as appropriate.

Results

Overall, 66 patients with 68 cystic lesions were identified during the study period. Eleven postnatally confirmed ovarian cysts were excluded from the data set, as well as 3 suspected ovarian cysts that resolved in the prenatal period and were assumed to have been correctly identified (accurate prenatal classification was confirmed by site data, Fig. 1). Therefore, after applying the study inclusion and exclusion criteria, 22 patients with 24 non-ovarian simple cystic lesions were reviewed (Fig. 1).

Overall, 67% of the cystic lesions resolved spontaneously either in the prenatal period (46%, $n = 11$) or with-

Table 1. Clinical outcomes of simple, non-ovarian cystic lesions

Clinical outcome	Lesions	%	Prenatal cystic mass evaluation				Postnatal days until
			GA at US detection, weeks	US scans[#]	mean max. area, cm^{2a}	max. area, median, range, cm^2	
Prenatal resolution	11	45.8	24.0±4.8	2.0±1.4	5.5±6.7	4.9, 0.1–24.6	–
Postnatal resolution[b]	5	20.8	26.5±10.0	2.2±1.6	8.7±8.8	5.8, 3.3–24.2	215±192
Surgery	7	29.2	23.2±5.9	3.3±1.5	23.0±24.9	15.3, 3.1–58.5	53±76
Benign persistent	1	4.2	27.1	1	0.9	–	–
Overall	24	100	24.4±6.2	2.4±1.5	11.8±15.3	5.3, 0.1–58.5	–

Study population included 22 fetal subjects with $n = 24$ masses (2 patients with 2 masses), mean maternal age = 29.5±4.0 years, fetal gender = 11 males (50%). Data are presented as mean ± standard deviation. GA, gestational age; US, ultrasound. [a] One-way ANOVA: $p = 0.072$, not significant at $\alpha = 0.05$. [b] Confirmed radiologic resolution with postnatal imaging.

in 18 months of life (21%, $n = 5$); the remaining patients required postnatal surgical intervention (29%, $n = 7$) except for 1 persistent benign cyst (3%, $n = 1$) that was managed conservatively (Table 1). In general, the cystic lesions that resolved had smaller maximum areas during their prenatal course (5.5 ± 6.7 cm^2 prenatal resolution, 8.7 ± 8.8 cm^2 postnatal resolution) than those that required postnatal surgery (23.0 ± 24.9 cm^2), but this difference did not achieve statistical significance (Table 1, one-way ANOVA: $p = 0.072$).

Fourteen of the 24 cystic lesions in our data set received a nonspecific diagnosis prenatally based on the US evaluations (Fig. 2). Of the 10 lesions suspected to be involved with a solid organ, 8 had suspected renal or suprarenal involvement and 2 had hepatobiliary involvement. The confirmed postnatal diagnosis of the cysts that were unresolved after birth is described in Figure 3.

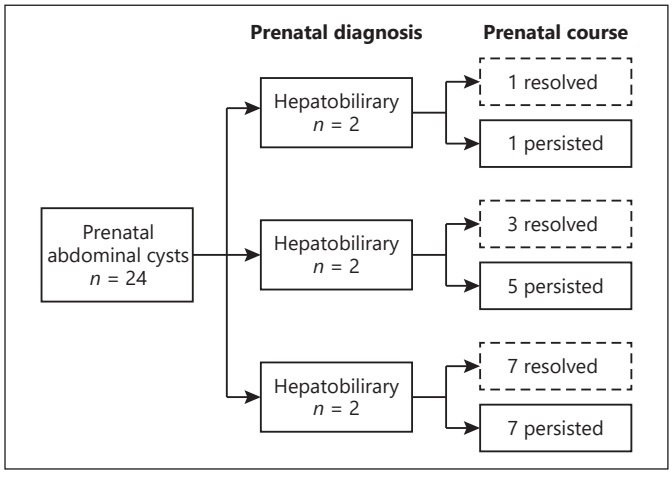

Fig. 2. Prenatal non-ovarian abdominal cystic lesions.

Discussion/Conclusion

Detection of fetal abdominal cysts with ultrasonography is fairly common and straightforward, but differential diagnosis for simple non-ovarian cystic lesions and prediction of clinical outcomes such as spontaneous resolution versus neonatal intervention are challenging [7, 9]. In addition, there is a paucity of data and wide variability in the reported rates of spontaneous resolution specifically for non-ovarian simple cystic lesions. At our center, the majority of non-ovarian cystic lesions had a nonspecific prenatal diagnosis, and 67% resolved in the perinatal period.

Unfortunately, in this small data set we did not identify a statistically significant size threshold that could accurately differentiate which cystic lesions would resolve and which would need surgery. This is likely because both resolution groups contained 1 larger lesion (>24 cm^2). In addition, several lesions with relatively small areas required surgical intervention, including a type 3 sacrococcygeal cystic teratoma (5.3 cm^2), type 1 choledochal cyst (3.1 cm^2), and pancreatic cyst (4.7 cm^2). Therefore, there was no absolute size threshold or gestational growth patterns in this small data set that effectively predicted spontaneous lesion resolution. Also, unlike ovarian cysts which are theorized to spontaneously regress in the absence of maternal hormonal stimulation, the mechanism of resolution of non-ovarian cystic lesions is yet to be elucidated. While it is tempting to speculate as to the varying etiology for resolution, there would be no scientific evidence to support these conjectures.

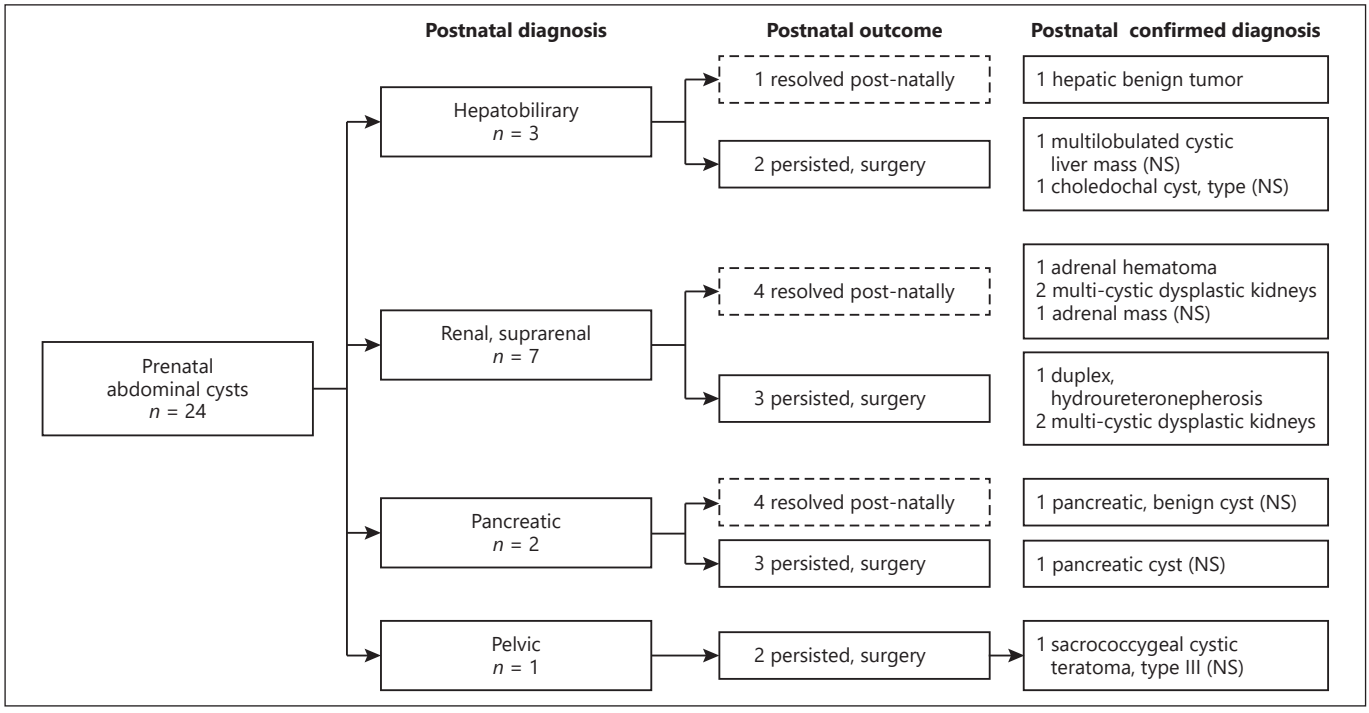

Fig. 3. Postnatal diagnosis of persistent cysts. NS, nonspecific prenatal diagnosis.

Table 2. Summary of prenatally discovered non-ovarian, simple, abdominopelvic masses that resolved before birth

Publication	Year	Prenatal abdominal mass diagnosis				
		total masses	urinary, suprarenal	hepatobiliary	gastrointestinal	nonspecific
Eyerly-Webb et al.	2020	11 of 24 (46%)	3 of 8 (38%)	1 of 2 (50%)	–	7 of 14 (50%)
Sanna et al. [5]	2019	10 of 24 (42%)	0 of 2 (0%)	2 of 5 (40%)	6 of 15 (40%)	2 of 12 (16%)
Husen et al. [8]	2019	5 of 47 (11%)	–	–	–	–
Catania et al. [2][a]	2016	1 of 22 (5%)	–	–	1 of 9 (11%)	–
Thakkar et al. [6]	2015	6 of 78 (8%)	2	–	–	4
Ozyuncu et al. [7]	2010	9 of 26 (35%)	–	–	–	–

All studies were retrospective reviews; data in this table were calculated for non-ovarian simple cystic masses (study inclusion criteria) based on the data available in the articles [2, 5–8]. [a] This study excluded retroperitoneal, urinary, thoracic, and spinal origin cysts.

There are several reports that describe the diagnosis and management of cystic or solid abdominal and pelvic prenatal masses, but studies have typically focused on improvement of diagnostic sensitivity rather than clinical prognosis [2, 5–8]. When comparing our prenatal resolution data to the most recent publications on this topic, our overall resolution rate specifically for non-ovarian simple cystic lesions was in the range of other studies in the literature (Table 2). However, there appears to be high variability in percentage of lesions that resolved prenatally in each study, and in many of these articles, prenatally suspected organ involvement was not reported. At our site, organ involvement could not always deliver a definitive diagnosis based on prenatal US imaging alone. If a definitive diagnosis was not possible based on imaging, our practice has been to follow up these lesions and increase the use of fetal magnetic resonance imaging (MRI) if clinically pertinent. In these instances, we used the terminology of nonspecific diagnosis, which has been previously used in the literature. Notably, other studies also had high percentages of nonspecific prenatal lesions as was seen at our center [5, 6].

Advanced US technology has improved greatly in the last decade, and the enhanced resolution facilitates the discovery of smaller and smaller fetal cysts. Therefore, it is becoming even more important to differentiate which cyst may become clinically problematic, and when there is no cause for alarm. Fetal cystic mass location and solid organ involvement are likely critical parameters in the prognostication; however, as was seen in this study, cyst specification with ultrasonography can be difficult and imprecise likely due to limited depth resolution, field of view, fetal motion, and viewing angles. MRI technology has been recently reported as a valuable adjunct to prenatal US in fetal diagnosis, and will likely prove a vital tool for more accurately determining lesion location, solid organ involvement, and pathology [1, 10–12].

The small study cohort and retrospective study design were the biggest limitations in this study. In addition, a large number of cystic lesions were designated nonspecific, making it difficult to draw meaningful conclusions from the location and solid organ involvement. In the future, a more detailed survey of lesion growth and evolution throughout gestation as well as enhanced localization with MRI could provide a more precise prenatal diagnosis. Ideally, each lesion type would also be described individually based on the unique origin and pathology. Future studies should assemble a multicenter data set based on a consistent monitoring protocol to better characterize the natural history and patterns of resolution for simple non-ovarian cystic lesions as relates to lesion size, origin, and solid organ involvement. These parameters, in addition to the timing of appearance during gestation, would likely be of interest for predicting clinical outcomes. Such data could inform a risk stratification system for these types of lesions and inform guidelines for prenatal management, including optimized prenatal imaging protocols and counseling.

Statement of Ethics

The research complies with the guidelines for human studies and was conducted ethically in accordance with the World Medical Association Declaration of Helsinki. The study protocol has been approved by the IRB at Children's Minnesota (#1703-044, approved March 30, 2017) and Allina Healthcare (#1053954, nonengagement determination, June 26, 2017, cede to Children's Minnesota). The IRB granted a waiver of informed consent. For each subject reviewed, a general consent was granted for release of data for research purposes.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

Eyerly-Webb: statistical analysis and manuscript writing, preparation, and approval. Joshi: data collection and manuscript preparation and approval. Lillegard: manuscript conception, data review, preparation, and approval. Dion: data collection and manuscript approval. Snowise: manuscript conception, preparation, and approval. Feltis: manuscript conception (PI), data review, preparation, and approval.

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