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ORIGINAL ARTICLE

Post-natal outcomes of antenatally diagnosed intra-abdominal cysts: a 22-year single-institution series

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Abstract
Purpose The aim of this study was to establish the post-natal diagnosis and outcome of antenatally diagnosed intra-abdominal cysts between 1991 and 2013 at our institution.
Methods All antenatally diagnosed intra-abdominal cysts between 1991 and 2013 were identified using a foetal anomaly database. The cysts were monitored for resolution. In all cases where the cyst had not resolved antenatally, additional post-natal scans were conducted. Antenatal diagnosis, post-natal diagnosis and outcomes were also recorded.
Results 118 cases of antenatal intra-abdominal cysts were identified over the 22-year study period with a 98 % live birth rate. The overall accuracy of an antenatal diagnosis at our institution was 92 %. 26 cases (22 %) resolved spontaneously in utero, the majority of which (77 %) were ovarian in nature. Four tumour cases were identified in the series, which included two neuroblastomas, one yolk sac tumour and one teratoma. 90 cysts persisted post-natally with 52 % requiring surgery. These primarily included choledochal and enteric duplication cysts as well as symptomatic solid organ cysts. Diagnostic revision was limited to 8 % of cases over the study period with an overall improvement over the last decade. Overall, 40 % of all antenatally diagnosed cysts required surgical intervention. In those cysts that persisted post-natally, 52 % required surgery.
Conclusions A fifth of prenatally diagnosed intra-abdominal cysts will resolve with most ovarian cysts regressing in utero. Half of all persistent cysts will, however, require surgical intervention. These data are useful for prenatal counselling and demonstrates the important role played by the paediatric surgeon in the overall management of intra-abdominal cysts.

Keywords Antenatal · Intra-abdominal cysts · Post-natal outcomes

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Introduction

Abdominal cysts diagnosed antenatally represent both normal variants and pathological entities. Antenatal counselling gives the paediatric surgeon the ideal opportunity to discuss these possibilities with prospective parents. The most common imaging modality used for antenatal scanning is ultrasound (US). The sensitivity and specificity of US were recently reported to be 88.1 and 95.7 %, respectively [1]. An accurate diagnosis is essential not only for accurate counselling but also for arranging the antenatal and post-natal care that may be required in the immediate newborn period. In 2008, our centre reported the outcomes of all antenatally diagnosed intra-abdominal cysts over a 13-year period. We sought out to extend this to a 22-year period and report the largest such series providing both all antenatal specialists with data that can facilitate counselling and provision of services.

Materials and methods

A retrospective case series review was conducted for all infants with antenatally diagnosed intra-abdominal cysts between 1991 and 2013. Cases were identified using the Congenital Anomaly Register for Oxfordshire, Berkshire and Buckinghamshire (CAROBB) at the John Radcliffe

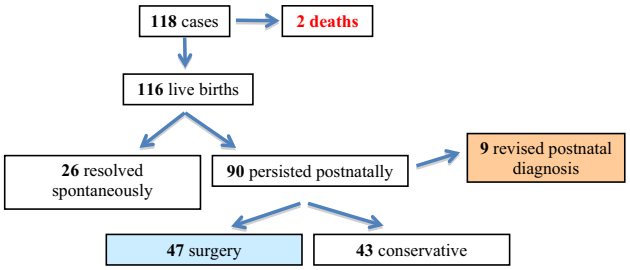

Fig. 1 Post-natal outcomes of all intra-abdominal diagnosed cysts

Hospital, Oxford, USA. Patient demographics and outcomes for all cases were recorded from departmental databases and medical records.

Antenatal screening protocol

As per the current UK National Screening Committee guidelines, all pregnant women in the region were invited for a 20-week anomaly scan at their booking hospital. All scans were performed by a trained sonographer who directly referred the patient to a tertiary foetal medicine specialist if any anomaly was found. All patients with a suspected surgical anomaly including intra-abdominal cysts were then reviewed in a joint Fetal Medicine & Surgical clinic accordingly. The frequency of scanning between 20 and 30 weeks gestation was four weekly. Thereafter, if the cyst still persisted, the frequency was increased to two weekly.

Post-natal scanning and management protocol

Post-natally, all infants had an US scan to check for cyst resolution and/or confirm the diagnosis within the first week of life. A surgical approach was taken for any infants who became symptomatic or were at high risk of complications due to the nature of the cyst. Similarly, if any cysts continued to enlarge or became complicated with rupture/bleeding, surgical management was again advocated. For all remaining asymptomatic cysts, routine follow-up in clinic was arranged until resolution of the cyst.

Results

118 cases of antenatal intra-abdominal cysts were identified over the 22-year study period with a 98 % live birth rate (two intra-uterine deaths, Fig. 1). The two deaths were patients between 1991 and 1993 with a post-natal diagnosis of cystic hygromas in the chest and the neck with associated foetal hydrops.

26 (22 %) resolved spontaneously in utero with normal post-natal scans. Of these, 20 (77 %) were antenatal ovarian cysts with the remaining comprising a suspected biliary cyst, urachal cyst and non-specific intra-abdominal cysts.

Table 1 Diagnosis of all surgically managed post-natal intra-abdominal cysts

Diagnosis	Frequency (%) $N = 47$
Choledochal	15
Enteric duplication	14
Meconium pseudocyst	5
Pelvic hydrometrocolpos	4
Mesenteric	3
Omental	1
Splenic	1
Renal	1
Ovarian teratoma	1
Complex enlarging ovarian cyst (8 cm)	1
Patent urachus	1

Table 2 Revised diagnoses from post-natal scans for antenatally suspected abdominal cysts

Initial diagnosis	Revised diagnosis
Ovarian cyst	Yolk sac tumour
Ovarian cyst	Meconium pseudocyst
Ovarian cyst	Enteric duplication
Choledochal cyst	Gastric duplication
Choledochal cyst	Non-specific
Mesenteric/duplication cyst	Meconium peritonitis
Mesenteric cyst	Non-specific
Mesenteric cyst	Non-specific
Meconium pseudocyst	Hydrometrocolpos with associated cloaca

90 (78 %) cysts persisted post-natally and of these 47 (52 %) required surgery (Table 1). The remaining 43 cases were monitored in clinic and either resolved spontaneously or remained asymptomatic. The majority of these were ovarian cysts; five of which resolved on serial post-natal scanning and nine of which persisted. Four tumour cases were identified in this series; two neuroblastomas, one yolk sac tumour and one teratoma. Antenatally all of these were suspected to be either adrenal or ovarian cysts but post-natally further imaging and biochemical testing confirmed tumours, which were subsequently managed by the oncology team.

The most common post-natal diagnosis requiring surgery was that of a choledochal cyst followed by enteric duplication and meconium pseudocysts. The majority of ovarian cysts in our series were managed conservatively but two cases needed surgical excision (one teratoma and

an enlarging complex 8 cm cyst). There were two other solid organ cysts (splenic and renal), which were symptomatic. The renal cyst was associated with hydronephrosis and required surgery for obstructive uropathy whereas the splenic cyst was excised due to associated bleeding.

In nine cases (8 %), the initial attributed diagnosis was revised at the post-natal scan giving an accuracy of 92 %. Table 2 illustrates both the suspected and revised diagnosis for these cases.

Overall, 40 % of all antenatal diagnosed cysts required surgical intervention with the remaining cases either resolving post-natally or observed remaining asymptomatic.

Discussion

Antenatal intra-abdominal cysts are heterogeneous lesions; the natural history of which depends on the type of lesion. The accuracy of antenatal diagnosis has implications both for effective counselling of parents as well as planning of post-natal management. As per the current UK National Screening Committee guidelines, a detailed antenatal anomaly scan is offered to all pregnancies between 18 and 21 weeks gestation. Any structural anomalies detected at this stage by a trained sonographer are then referred to a foetal medicine specialist at our institution including scans from surrounding district general hospitals. All such anomalies are then recorded by our centre in the Congenital Anomaly Register for Oxfordshire, Berkshire and Buckinghamshire (CAROBB).

Our centre serves a local population in excess of three million with the total number of births (live and stillbirths) within the Thames Valley gradually increasing over the study period from 28,966 between 1991 and 1995 to 37,729 between 2006 and 2010 (birth rate approximately 12/1,000–16/1,000) [2]. There has been no significant change in the ethnic composition of our population in the last 20 years. The total notifications made prenatally with a confirmed anomaly at birth have, however, remained stable at approximately 40 % of all notifications during our study period. Similarly, the termination of pregnancy rate has also remained stable with approximately 43 % of prenatally diagnosed confirmed cases resulting in termination.

The most common diagnosis in our study was that of an ovarian cyst (29 % of all cases). These findings are consistent with published literature reporting ovarian cysts to be the most common cystic lesion in the neonatal period [3]. Almost 2/3 of these cysts in our case underwent spontaneous resolution in utero which is higher than that has been reported. In a recently published series of 29 suspected antenatal ovarian cysts, 20 were confirmed post-natally and of these 35 % had undergone spontaneous regression in utero [4]. Most ovarian cysts that persist are asymptomatic as was found to be the case in our study. However, larger cysts usually greater than 5 cm are at risk of torsion, and therefore may advocate surgical excision [5]. One such complex 8 cm cyst was excised in our series due to its size and associated features of haemorrhage on US imaging.

Overall half of all cysts that persisted post-natally required surgical intervention. Most commonly encountered were choledochal and enteric duplication cysts. Choledochal cysts in the neonatal period usually arise from an intrinsic weakness in the biliary tree presenting with focal biliary dilatation [3]. There is also a strong association between choledochal cysts and biliary atresia [3]. Complications that may arise from these lesions include ascending cholangitis, rupture, gastric outlet obstruction, cholestasis, hepatic cirrhosis and portal hypertension [6]. Surgical management of these cysts is, therefore, advocated and is likely to explain the highest frequency of cases that required surgical excision in our series.

This series is consistent with our previously published data between 1991 and 2004, where the most commonly excised cysts remained choledochal and enteric duplication cysts with most ovarian cysts resolving [7]. Over the next decade as detailed above, the birth rate and termination frequency had not change significantly and this may in part explain the similar frequency with which we observed these cysts and their subsequent surgical and conservative management.

Neonatal malignancies are extremely rare and are found in 1/12,500–27,500 live births accounting for 2 % of all childhood cancers [8]. Neuroblastomas are the most frequently occurring neonatal tumours; the majority of which are suprarenal in nature as observed in our series. The prognosis is generally favourable and follow-up is advocated to assess for spontaneous regression. In this study, four tumour cases were identified; two of which were found to be neuroblastomas. Both cases were initially managed conservatively and followed up by the oncology team. Two ovarian neoplasms were also identified; yolk sac tumour managed by oncology and a >4 cm teratoma that was excised. Ovarian neoplasms also occur infrequently in the neonatal period but reported cases most often describe stromal tumours [8].

Antenatal US scans are becoming increasingly more sophisticated in the detection of congenital anomalies. US scans offer the advantage of attaining high-resolution images without the need for ionising radiation. Whilst technical advances are rapidly occurring in US imaging, it is still challenging to accurately distinguish between various different antenatal lesions [3]. The overall detection rate of antenatal cysts was 5.4/year. Between 1991 and 2004, this was 4.2/year; whereas over the last decade, this figure has risen to 7/year. Over, the same time period,

between 1991 and 2004, 11 % of cases were incorrectly diagnosed at our institution [7]. However, this figure has markedly improved over the last decade with a 5 % revision rate. These results can be explained by technical improvements in imaging as well the increasing experience gained over the years by our specialists. The cysts that had a revised diagnosed post-natally included both ovarian and choledochal cysts. Importantly, this demonstrated that antenatally these cysts may appear simple but post-natal imaging may reveal a more complex nature. This re-emphasised our policy of all cases receiving a post-natal scan to confirm the suspected antenatal diagnosis.

Our experience over the last 22 years is the largest published series to date. Overall, a fifth of cysts in our series resolved and of these, the majority were ovarian in nature. Of those that persisted, approximately half needed surgical intervention. This can provide a useful reference point for antenatal counselling and help guide further management; either conservative or operative.

With increasing local experience, this study demonstrates a high level of diagnostic accuracy that can be attained. We have also shown the importance of collaboration between foetal medicine and paediatric surgery required in the management of these lesions and advocate a close working relationship between these two specialties. Despite this being a single-centre study, the size of the cohort established, allows us to adequately demonstrate the natural history of antenatally diagnosed intra-abdominal cysts.

Conflict of interest Nil.

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