

Management of adnexal cysts detected in the antenatal period: review

Abstract

The aim of this review is to increase the understanding of management of adnexal cysts detected antenatally. Advancements in ultrasound technology have aided in the early detection of adnexal cysts. Adnexal cysts are recognized as most often being a consequence of the hormonal milieu associated with pregnancy. Often, these cysts are simple-appearing and measure smaller than four centimeters. However, these cysts can be large and may lead to adnexal torsion and hemorrhage. This article will provide a summary of the literature published in regards to management of neonatal adnexal cysts.

Keywords: adnexal, ovarian, neonatal, cysts

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Introduction

Owing to dramatic improvements in ultrasound technology, adnexal cysts can be detected antenatally.¹ Adnexal cysts are the most common intra-abdominal cystic lesion found in female newborns, constituting about 10% of all abdominal cysts in neonates.² Adnexal cysts often form in response to the hormonal milieu associated with pregnancy.³ Small physiologic follicular cysts are seen in approximately 1 in 2500 of live births.⁴ Often, these cysts are small and simple-appearing and resolve spontaneously. However, some of these cysts are large and may lead to adnexal torsion and/or hemorrhage. Management of neonatal adnexal cysts is controversial but should be aimed at ovarian preservation.

Development of the ovary

Prior to assessing the optimal approach to managing neonatal adnexal cysts, it is paramount to understand the mechanisms that lead to the formation of these masses. Ovarian development begins at 6 weeks of gestation with rapid mitosis that increases the number of oogonia from 10,000 to 600,000 by 8 weeks. By 20 weeks of gestation, the number of oogonia increases to 6-7 million. Rapid follicular atresia, along with follicular growth and development, then occurs subsequently leading to 1-2 million germ cells remaining at the time of birth.⁵ Follicular cyst development has been documented to occur as early as 28 weeks of gestation.⁶ By the end of pregnancy, preantral follicles and antral follicles with granulosa cell and theca cell differentiation are normally present. This hormonal interplay continues well on into the reproductive period as adnexal cyst formation coinciding with anovulation is very common in the adolescent period.⁷

Diagnosis of antenatal and neonatal adnexal cysts

Advancements in obstetric ultrasonography have permitted clinicians to detect adnexal cysts antenatally that would have otherwise gone unnoticed. However, adnexal cysts are not always easy to discern from other structures. Therefore, it is essential that adnexal cysts be differentiated from mesenteric and omental cysts. Thus, the diagnosis of adnexal cyst is based on the four following criteria: (1) female gender, (2) non-midline regular cystic structure, (3) normal-appearing urinary tract, and (4) normal appearing gastrointestinal tract.⁸ Inability to adequately diagnose an abdominal mass on ultrasonography presents a dilemma in management.

As mentioned prior, adnexal cysts comprise the majority of abdominal masses in the female neonate. Furthermore, at birth up to 98% of female neonates have small physiologic adnexal cysts detected on ultrasonography.⁹ Cysts tend to occur secondary to ovarian hyperstimulation by placental chorionic gonadotropin and maternal hormones¹⁰ and carry the appearance of a hypoechoic structure. Internal echoes, if present, are most likely a reflection of hemorrhage or organized hematoma. Rupture of a neonatal adnexal cyst is a rare occurrence.¹¹ These cysts are also often unilateral, thin-walled and unilocular.¹²

Management of antenatal and neonatal adnexal cysts

To date, no randomized trials have been performed assessing the optimal management of adnexal cysts detected antenatally. Management approaches have been based on the multitude of case reports and series that have been published. For the purposes of this article, a PUBMED Search using the search words, "adnexal," "cysts," "ovarian," "antenatal," and "neonatal," were used to extract articles focusing on this condition. This article will provide a comprehensive review.

Expectant management

With the introduction of ultrasound for routine monitoring during pregnancy, the incidence of adnexal cysts detected has increased but there has been some elucidation as to the natural course of these cysts. Some have reported that cysts that are smaller than four centimeters tend to spontaneously resolve over the span of three to four months.¹³ while others have noted that resolution may be expected by about six months of age given the cessation of passage of placental human chorionic gonadotropin and maternal hormones.¹⁴ In a study of 16 adnexal cysts in 16 fetuses, 7 of the 11 adnexal cysts that were simple-appearing disappeared on follow-up imaging during pregnancy or within two months of birth.¹⁵ These findings were confirmed by a larger study of 82 fetal adnexal cysts in 79 patients that were followed sonographically for several months in which 27 out of 55 simple adnexal cysts remained simple in appearance; median size was four centimeters.¹⁶ Based on this evidence, conservative management appears to be favored for those cysts measuring four centimeters or less. However, there is controversy regarding monitoring of adnexal cysts detected antenatally that measure larger than four centimeters given the risk of adnexal torsion and the rare, but reported occurrence of auto-amputation of the adnexae. Auto-amputation is characterized

by either a free-floating tubo-ovarian remnant or adhesion of the structure of other pelvic structures as opposed to being in its usual anatomic location and is believed to result from chronic ovarian torsion leading to devascularization.^{17,18} Published articles on auto-amputation of the adnexae have been limited to case reports.

In a study of 67 adnexal cysts detected antenatally, 18% of the masses were complex on initial scan. On follow-up imaging, 55% were complex.¹⁹ Neonatal surgery was performed for 64 cysts of which 56% had undergone torsion, indicating that adnexal cysts do confer a risk of ovarian loss. In an effort to accurately target those with adnexal torsion, some have theorized that certain ultrasonographic markers may be predictive of this condition. Chinchure et al.²⁰ suggested that the presence of internal echoes with a “fish-net appearance,” fluid-debris level and cysts with an echogenic nodule were sonographic markers highly suggestive of adnexal torsion. However, adnexal torsion continues to be a difficult diagnosis to make preoperatively. Surgical management, when warranted, also continues to be a controversial topic.

Laparotomy

Historically, surgical management of adnexal torsion consisted of laparotomy. In a case series by Widowson et al.²¹ published in 1988, 7 newborns with adnexal cysts detected antenatally underwent laparotomy soon after birth. In 3 of the cases, salpingo-oophorectomy was performed while cystectomies and/or enucleation were performed in the other 4 cases. Pathology for these 4 cases was benign and consistent with follicular adnexal cysts and normal ovarian tissue. This study showed a shift from salpingo-oophorectomy to one aimed at ovarian preservation but also showed that in cases in which bilateral ovarian cysts were present, surgical management carries a high risk of rendering a patient sterile. They acknowledged that cysts measuring less than four centimeters were most likely to undergo spontaneous resolution but addressed the dilemma associated with cysts measuring larger.

In 1996, Mas et al.²² reported a case of a woman whose routine ultrasonographic imaging at 32 weeks gestation revealed a five centimeter mass consistent with an adnexal cyst in the female fetus. Imaging five weeks later indicated that the cyst had become larger and fluid debris was appreciated. A cesarean delivery was performed 24 hours later. The newborn underwent a laparotomy and a unilateral salpingo-oophorectomy was performed. These authors also acknowledged the difficulty faced in attempts to salvage the ovary that is associated with large, complex cysts.

In a larger case series, Mizuno et al.²³ published their findings on 17 patients between 1983 and 1997 who presented with neonatal adnexal cysts, of which all but 2 were detected antenatally. 15 cases were diagnosed during routine antenatal ultrasound examinations from 30 to 37 weeks of gestation while the other 2 were diagnosed postnatally due to palpable abdominal masses and distention. In one patient, the cyst spontaneously resolved a few days after birth while the other 16 newborns underwent laparotomy within 1 month of birth. The size of the cysts ranged from 23 – 100 mm. Out of 16 patients, 9 had adnexal torsion and 7 out of these 9 showed necrosis in the ovary and oviduct. Adnexal cystectomies were performed in the remaining 7 patients. Salpingo-oophorectomy was performed in 5 of the patients who had adnexal torsion.

Percutaneous aspiration

Given the complications associated with surgery, the option of percutaneous aspiration began to be explored. Some authors advocated

for this course of management as a means of not only reducing the risk of adnexal torsion but also that of preterm labor, chorioamnionitis and polyhydramnios secondary to the presence of large, expanding cystic formations.¹⁹ In a case report by Shozu et al.²⁴ an ultrasound scan revealed an adnexal cyst in the fetal lower abdomen at 29 weeks of gestation that measured 6.5 centimeters at 33 weeks of gestation. The female infant was delivered vaginally at 41 weeks of gestation without any complications. Transabdominal needle aspiration of the cyst fluid was performed on day fourteen of life. Cytology of the cyst fluid was negative. Follow-up until the baby was a year old showed no recurrence of the cyst. Similar findings were reported in a study by Meager et al.²⁵ in which hemorrhage into the fetal adnexal cysts was suggested on ultrasound and in which percutaneous needle aspiration under ultrasound guidance was performed successfully, obviating the need for surgery. Another case series of 3 patients²⁶ provided data supporting percutaneous needle aspiration as a viable option. In all 3 patients, surgery was avoided and the cyst aspirate was confirmed to be ovarian in origin as based on fluid biochemistry that showed high amounts of estradiol, progesterone, and testosterone. An even larger case series of 13 patients²⁷ also attested to the safety of percutaneous aspiration given the lack of maternal and/or fetal complications. Additionally, 9 of these 13 patients went on to have a vaginal delivery. However, in the Mizuno study, two patients underwent percutaneous needle aspiration under ultrasonographic guidance but the cysts that were drained reverted to their previous size a few days following aspiration. Furthermore, a case report by Pulgandla and Laberge²⁸ documented a lethal outcome after percutaneous aspiration of a presumed adnexal cyst. Percutaneous needle aspiration was atraumatic and revealed serous fluid with a high estradiol level. However, surgery was subsequently indicated for clinical deterioration and a midgut volvulus with extensive necrosis was found secondary to a jejunal duplication cyst. Management of adnexal cysts via percutaneous aspiration continues to be controversial.

Laparoscopy

As techniques in laparoscopic surgery became more refined in the adult population, questions were being raised as to its applicability in the pediatric and neonatal population. In 1995, van der Zee et al.²⁹ described two cases of laparoscopic-assisted cyst mobilization with resection through 1 centimeter lower abdominal incisions in the newborn population. In a case report by Decker et al in 1999, a three-port technique was used with 5 millimeter skin incisions.³⁰ However, in spite of the agreement among surgeons that laparoscopy allows for a minimally invasive approach to management of neonatal adnexal cysts, there was concern for tolerance of pneumoperitoneum in the newborn and restrictions on operating space given the small abdominal cavity.³¹ However, now, laparoscopy is considered to be the ideal method in managing neonatal adnexal cysts as based on case reports published over the past 20 years. From 1996-2001, Tseng et al.³² identified six patients with perinatal torsion of an adnexal cyst and subsequently performed laparoscopy on all six patients. A two-port technique was used which involved obtaining adequate pneumoperitoneum with a Veress needle and placement of a 5 millimeter port in the infraumbilical position. The second port was placed in either the left or right lower midabdomen as determined by which ovary was affected. There were no complications stemming from the procedure and five newborns were discharged the day after surgery. A more recent case series by Schenkman et al.³³ studied outcomes from 20 patients who underwent surgery via laparotomy (n = 8), traditional laparoscopy (n = 5) and laparoscopic-assisted transumbilical extracorporeal cystectomy (LATEC, n = 7). LATEC is initially performed in the same manner as traditional laparoscopy but

at the termination of the procedure, the incision that was made at the umbilicus is extended from 5 to 15 millimeters in order to facilitate removal of the cyst. Postoperative courses were similar between those patients who underwent laparotomy versus traditional laparoscopy but those patients who underwent LATEC experienced the shortest operative times and cosmetic results were outstanding, providing an option to those who find difficulty in performing laparoscopy in a confined space. A more recent case series by Marinković et al documented outcomes of 13 newborns³⁴ who presented for surgical treatment of ovarian cysts; 8 newborns underwent laparotomy while 5 underwent laparoscopy. No complications were encountered and these authors also concluded that laparoscopy is as safe and effective as classical laparotomy in the management of neonatal adnexal cysts. Hence, studies do support the use of a minimally invasive approach when performing surgery for suspected torsion and/or hemorrhage in the neonatal population.

Conclusion

Neonatal adnexal cysts are common and are frequently detected on routine prenatal ultrasonography. The majorities of neonatal adnexal cysts are benign and resolve spontaneously. Expectant management is favored for those simple-appearing adnexal cysts that measure four centimeters or less. However, a small percentage is large and/or complex and may lead to ovarian loss. Based on the existing evidence, a minimally invasive approach appears to be favored when faced with large, complex cysts or when adnexal torsion or cyst rupture is suspected. However, the role of timing of percutaneous aspiration or surgical intervention, in the presence of a high suspicion of adverse sequelae, is controversial. Current evidence on management of neonatal adnexal cysts continues to be confined to case series.

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Conflicts of interest

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