

Complex Neonatal Ovarian Cyst: A Retrospective Study from Pediatric Institute

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Abstract

Background: Majority of neonatal ovarian cyst spontaneously regress within one year of age. Thus, most of the surgeons favor “wait-and-see policy” while dealing with such case. Surgical intervention is reserved for complicated and persistent ovarian cyst.

Objective: Scrutiny over the clinicopathological behavior and surgical management of complex neonatal ovarian cyst.

Material and Methods: Between March 2013 and February 2018, seven cases of unilateral, complicated neonatal ovarian cysts were managed in our institute. The data of clinical presentations, perinatal investigations, preoperative resuscitation, timing and indication of surgery, perioperative management and post op follow-up notes were collected, tabulated and analyzed.

Results: All seven cases required surgical intervention. Five patients underwent operation in neonatal period and two patients in post neonatal period. Emergency surgery was undertaken in four cases because of torsion, intracystic hemorrhage and extraordinary large cyst.

Conclusion: Acute life-threatening complications may develop early in neonatal period even in previously asymptomatic neonatal ovarian cyst. Thus, a close monitoring and prompt surgical intervention are imperative while dealing of such cases.

Keywords: Neonatal Ovarian Cyst (NOC); Surgical Intervention; Complex Neonatal Ovarian Cyst (CNOC)

Introduction

Simple follicular ovarian cyst is the most commonly encountered cyst in female fetuses/newborn which usually involute within first year of life. Fetal ultrasonography (USG) at 3rd trimester usually detect a cystic lesion which must be differentiated from other intra-abdominal cystic lesion; simple renal cyst, multi-cystic dysplastic kidney (MCDK), Hydronephrosis, Urachal cyst, hydrocolpos, enteric duplication cyst, meconium pseudocyst and choledochal cyst [1]. A multi-cystic cyst or cyst more than 2 cm in size may increase in size and

get complicated. Thus, a close monitoring with serial USG is necessary in order to detect complications and prompt surgical intervention. Sometimes, contrast enhanced computed tomography (CECT) is necessary to clear the preoperative diagnostic dilemma. Theoretically, multiple treatment options for neonatal ovarian cyst (NOC) are well described in literature (aspiration, observation and laparoscopy) but, open surgery is the only viable option for the management of complex neonatal ovarian cyst (CNOC) especially in acute torsion and intracystic hemorrhage.

Material and Methods

We performed a retrospective study with 7 cases of NOC who were treated in the department of pediatric surgery of our institution over a period of five years. The patients who were operated for CNOC are included in this study. We excluded the asymptomatic NOC, fortuitously observed NOC during laparotomy for other indications and incidentally encountered NOC by USG from our study. A diagnosis of CNOC was confirmed by USG. CECT was advocated only in doubtful cases. Blood hemogram, serum alpha-fetoprotein (AFP) and Beta human chorionic gonadotropin (hCG) were measured in all cases. The presenting symptoms, size and site of the cyst, associated malformations, perinatal medical management, indication and age at operation, types of surgical intervention, perioperative management and pathologic reports of the NOC were recorded, tabulated and analyzed (Table 1).

No	Age and weight	Presentation	Investigations and Postnatal findings	Indication and Age at Operation	Per-operative findings and procedure
1	1 day/ 2.7 kg	Non-progressive abdominal distention since birth.	Postnatal USG - Large cystic space occupying lesion (14 cm X 9 cm) with multiple septations. Complicated ovarian cyst.	Developed acute abdomen on 6 th day Emergency exploration was done on 7 th day.	Right sided ovarian cyst with intracystic hemorrhage. Right sided salpingo-oophorectomy done. Left Ovary - N
		Antenatal USG-hepatomegaly			
2	6 days/ 4.6 kg	Perinatal asphyxia with large baby according to gestational age (Approx. 46 wks).	Postnatal USG - cystic SOL (5.3 cm X 3.9 cm) in the midline of the abdomen. Neonatal ovarian cyst CECT needed to confirm the diagnosis. 24 hours VMA-WNL	A planned Surgery was done on 15 th day to excise cyst.	A cricket ball shaped right ovarian cyst with long pedicle. The cyst was excised along with right ovary and right fallopian tube was preserved Left Ovary - N
		Antenatal USG and Large cystic SOL in the lower abdomen (R) Hydronephrosis.			
3	2 d/ 2.2 Kg	Gradually increasing cystic mobile lump in lower abdominal region since birth	Postnatal USG - Peripherally enhancing cystic SOL extending from undersurface of liver to pelvis. Large ovarian cyst.	The baby developed symptoms due to pressure effects from huge intra-abdominal mass. Surgery was planned on 7 th day to excise the cyst.	Right sided ovarian cyst with intracystic hemorrhage. The cyst was excised with preservation of right fallopian tube and specimen sent for histopathological examination. Left ovary - N
		Antenatal USG and Mesenteric cyst			

4	4 d/ 2.5 Kg	Huge abdominal mass since birth.	Postnatal USG - cystic SOL (8.5 X 6.7) cm occupying whole of the abdomen which pushes the gut loops peripherally. Mild hepatomegaly was also described. Both the ovaries could not be seen separately.	The Patient developed respiratory distress due to mass effect. An Emergency surgery was performed for decompression on 8 th day.	Right ovarian cyst with intracystic hemorrhage. Right sided salpingo-oophorectomy done. Left ovary - N.
		Antenatal USG- Multi-cystic SOL MCDK			
5	7d/F/3.2 Kg	Swelling in the lower abdomen since birth.	Postnatal USG - intra-abdominal cystic (4.8 X 5.8) cm mass and only left ovary was visible.	A Planned surgical exploration was done on 14 th day.	Tennis ball shaped right ovarian cyst. Right salpingo-oophorectomy done. Left ovary - N
		Antenatal USG- NAD.			
6	14d/F/3.1 Kg	Lower abdominal fullness noticed by mother after 2 weeks of birth.	Postnatal USG - right sided ovarian cyst (4cm X 3cm). We planned to follow up the patient with serial USG. Serum AFP and beta HCG were WNL.	Patient came back after 2 weeks with huge abdominal lump with dragging pain. USG - intraabdominal complex SOL (13.5X9.6) cm, both ovaries could not be seen separately. Repeat AFP and beta HCG – WNL. A planned exploration was done.	Right sided complex large ovarian cyst. Right salpingo-oophorectomy done. Left ovary- N.
		Antenatal USG- NAD			
7	25d/ F/ 3.1Kg	Incidentally diagnosed ovarian tumor by local physician at the age of 25 th days of life.	Postnatal USG- right ovarian cyst (3.5 X 3.2) cm. AFP and beta HCG – WNL. We planned to follow up the patient. The patient was asked to attend in follow-up clinic monthly.	She came back after 3 weeks with abdominal lump and severe pain abdomen. CECT - abdomino-pelvic enhancing complex SOL. Suggestive of complex right sided ovarian cyst. An emergency exploration was done.	Torsion of right ovarian cyst with intracystic hemorrhage. Right salpingo-oophorectomy done. Left ovary- N.
		Antenatal USG- NAD			

Table 1: Clinico-pathological spectrum of neonatal ovarian cyst (n = 7).**Abbreviations**

USG: Ultrasonography; MCDK: Multi-cystic Dysplastic Kidney; NAD: No Abnormality Detected; SOL: Space Occupying Lesion; VMA: Vanillylmandelic Acid; WNL: Within Normal Limit; AFP: Alpha-fetoprotein; HCG: Human Chorionic Gonadotropin; CECT: Contrast Enhanced Computed Tomography; N: Normal.

Out of seven, three had developed acute abdomen because of torsion and intracystic hemorrhage where emergency exploration was done. Two cases were planned for operation as one had non-regressive NOC (more than 4 cm) and another had extraordinary large cyst (Figure 1). Other two patients had unilateral, uncomplicated NOC of less than 4 cm size. Thus, wait and see policy was considered for them. Unfortunately, both of them returned back in the emergency within 3 weeks with acute abdomen. One of them had torsion of the OC with intracystic hemorrhage and underwent emergency exploration after initial resuscitation (Figure 2). Another patient had developed huge abdomino-pelvic cystic mass. CECT abdomen suggested a large right sided OC (13.5 X 9.6 cm). After repeat beta hCG and AFP, surgical exploration was arranged. Postoperatively, these two cases were kept in ICU for ventilatory support and close monitoring. They were discharged on 5th and 6th post-operative day respectively.

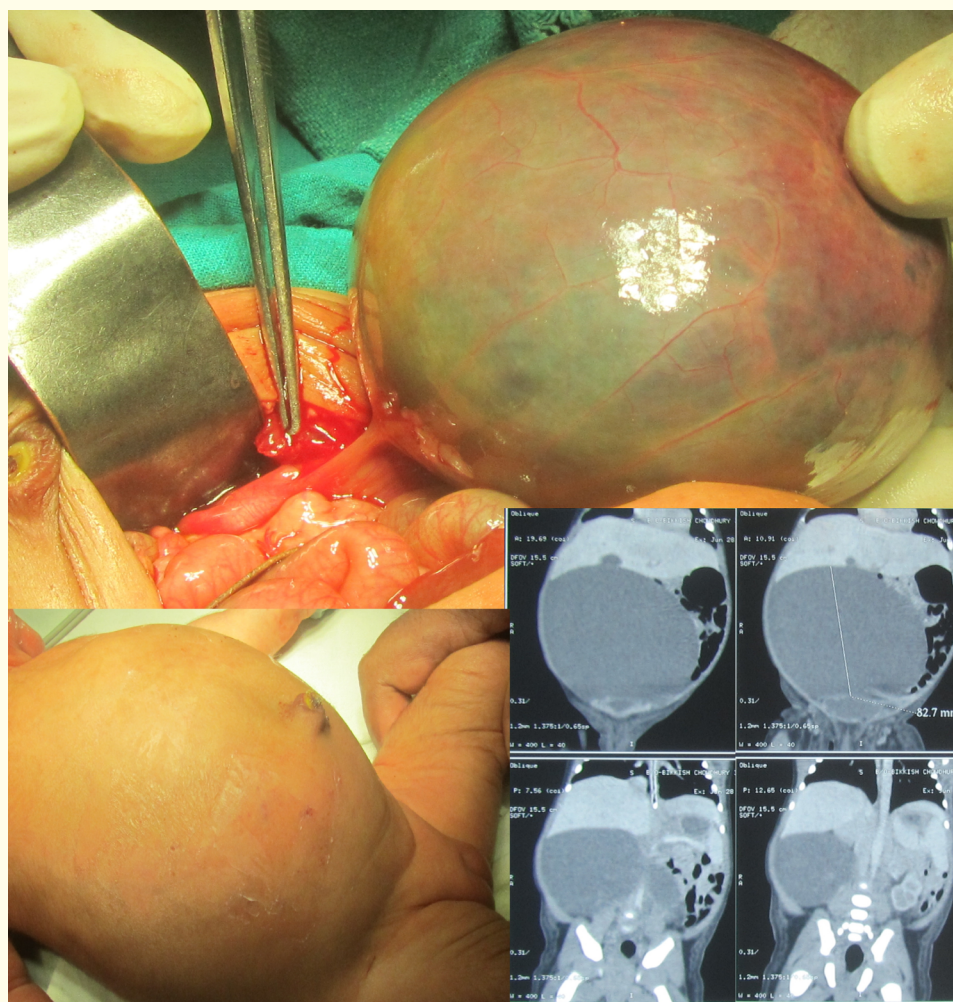


Figure 1: Perioperative photograph and Pictures of computed tomography show a large right sided ovarian cyst in a neonate on 7th day of life.



Figure 1: Perioperative photograph and Pictures of computed tomography show a large right sided ovarian cyst in a neonate on 7th day of life.

Results

In this study, all patients had right sided OC with normal left ovary. None of the patient had any evidence of intersex problem. Three (42.85%) patients had cystic mass in antenatal USG. Emergency exploration was done in four (57.15%) patients while three (42.85%) patients underwent planned surgery. Preoperatively, two (28.57) neonates were kept in SNCU, one for perinatal asphyxia and the other because of dyselektrolytemia. Operative intervention was undertaken for this two patients after initial medical stabilization. Torsion and intracystic hemorrhage were the immediate issue for emergency exploration. Five (71.42%) patients underwent unilateral salpingo-oophorectomy and two patients had oophorectomy. Five patient (71.42%) underwent operation in the neonatal period (mean age at operation was 10.2 days and range 7 - 15 days). The mean duration of post-operative hospital stay was 5 days, varying between 3 - 7 days. Two neonates were initially planned for wait and see management policy as they had uncomplicated NOC of less than 4 cm size. Both the cases ultimately needed surgical excision of the cyst after 2 weeks and 3 weeks respectively. USG failed to detect the OC in two cases (28.57%) and CECT was done in those cases. The biopsy reports revealed twisted follicular cyst in three (42.86%) cases and intra cystic hemorrhage in four (57.14%) cases. All patients are doing well on follow-up. None of the patient had any evidence of recurrence in post-operative period. There was no evidence of recurrence in any of the patient at 24 months follow-up.

Discussion

The first antenatal detection of OC was done in 1975 by Valenti. An antenatal USG after 28th gestational weeks easily diagnose the OC though, it can be detected as early as 19th week. It is postulated that fetal exposure to excessive gonadotropins increases the incidence of OC. A raised chorionic gonadotropin is often observed in complicated pregnancies with large placenta such as in diabetes, pre-eclampsia and Rh incompatibility [2]. Furthermore, fetal hypothyroidism and congenital adrenal hyperplasia may raise the incidence of OC. After birth, gonadotropin level in neonate gradually decrease and spontaneous regression of the OC occurs. A non-regressing/persistent NOC may be due to neonatal ovarian hyper stimulation because of immature gonadostat mechanism.

The clinical behavior of NOC depends on its size as most of the cysts are simple follicular type. A large fetal cyst may cause uterine dystocia, polyhydramnios and fetal pulmonary hypoplasia. After birth, it may present with pain, irritability, vomiting, tachycardia and intestinal and urinary obstruction due to pressure effects. Moreover, torsion, intracystic hemorrhage, cyst rupture with hemoperitoneum, auto-amputation of ovary, sudden infant death syndrome and incarceration of an inguinal hernia may occur. Torsion is the most frequent

complication (50% to 78%) followed by intracystic hemorrhage [3,4]. Torsion of the ovary can cause circulatory impairment that leads to hemorrhagic infarction. It is more common (20 - 32%) in the perinatal period than post-neonatal period. A large (greater than 5 cm) cyst with long pedicle is more prone to torsion. It has been postulated that torsion occurs more frequently in the right ovary than in the left ovary because of the close anatomic association of the sigmoid colon with the left ovary [5]. In our case series, torsion in the right ovary was found in five patients. Several management strategies like; USG guided aspiration, wait-and-see policy, laparoscopy and surgical excision have been described in literature. Each of the options needs to be selected carefully considering the size, duration, timing (pre-natal/postnatal), complications and progression of the cyst after birth. A large OC in the intrauterine period may affect the spontaneous vaginal delivery. In this situation, USG guided puncture may be helpful [6]. Kwak., *et al.* and Kessler., *et al.* performed prenatal US-guided aspiration of OC to avoid intrauterine torsion. They believe, US-guided aspiration (both before and after delivery) is safe, effective and repeatable procedure that preserves the ovarian tissue in a higher percentage than surgery [7]. However, the risk of recurrence is high. Moreover, prenatal intervention carries potential risks of cyst rupture, peritonitis, preterm labor, chorioamnionitis, and fetal injury. Many surgeons claim that, majority of the cysts resolve spontaneously completely after birth [8]. They recommend “wait-and-see policy” with serial USG up to one year especially when size of the cyst is less than 4 cm. Enriquez., *et al.* [9] reported 11 cases of NOC where all the cysts completely resolved at the mean age of one year. Similarly, Laessoe L., *et al.* reviewed NOC cases and concluded that NOC of less than 4 cm size should be followed up by serial USG up to one year. Mizuno M., *et al.* revealed that small simple ovarian cysts < 4 cm in diameter could be observed carefully with serial ultrasonography. Spontaneous regression occurs in more than half of NOC cases in the prenatal or post-natal period. However, long-term outcome and risk to future fertility is unknown. Practically, there is no precise guide for the monitoring and treatment of NOC below 4 cm. In our series, none of the cysts showed any evidence of shrinkage on serial USG. Moreover, five patients eventually developed complications in the form of intracystic hemorrhage and torsion. Laparoscopy for NOC provides both diagnostic and therapeutic purposes. Furthermore, visual assessment of the cyst allows multiple management options like; aspiration of the cyst, cystectomy, decapsulation of the ovary, stripping of cysts and oophorectomy [10]. However, laparoscopy in neonates is a really technically demanding operation and it is better to avoid in symptomatic and complicated cysts [11]. Complications (torsion and hemorrhage) of a cyst suspected when there is sudden changes in the size and development of symptoms in a previously asymptomatic cyst. Usually, USG can detect the complex nature of the cyst. Sometimes, CECT or magnetic resonance imaging (MRI) is needed to determine the age of the hemorrhage in large cysts [12]. In addition to that, they are used to exclude the other possible differential diagnosis. Surgical excision of the cyst is regarded as standard procedure till now. Indeed, it is the only option in patient with symptomatic large cyst, complex cysts and recurrent/persistent cysts which failed to resolve with all other minimally invasive procedure. Laessoe., *et al.* postulated that any postnatal symptomatic cysts or cysts with a diameter greater than 5 cm that do not regress or enlarge should be treated surgically [13]. In addition to that, early surgery may allow preservation of ovarian tissue where de-torsion is possible. The histopathological study of all torsed cysts revealed hemorrhagic infarcts. Malignant tumors are rare in the neonatal period, but benign cystic teratomas are the most common ovarian tumors [14]. Lymphangiomas also are counted among the hamartomas lesions of the fetal-neonatal ovary. However, in our study, all the cysts were benign follicular cyst without any malignant transformation. Practically, we did not encounter any significant differences between emergency and planned surgical intervention, even with an extraordinary large cyst. Truly, there is no consensus in the modality and timing of the treatment or monitoring of NOC. Considering our result and recent literatures, we vouch that specific approaches based on institutional practice and available resources always playing an important role in the management of NOC.

Conclusion

As far as our small series is concerned, surgery may be the most important tool in the management of symptomatic, complex and extraordinary large NOC. A delay in the management increases the risk of torsion, intracystic hemorrhage acid-base disturbances, sepsis and so also the mortality. However, prognosis and the long term outcomes of unilateral NOC are satisfactory as most cases are benign follicular cyst which has least chance of recurrences after complete removal.

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Conflicts of Interest

Nil.

Authors Contribution

Pankaj Halder: First Author, Literature search and Editing of manuscript.

Arpita Das: Literature search.

Kartik Chandra Mandal: Concept and design.

Dipanwita Mitra: Literature search and manuscript edition.

Bidyut Debnath: Editing of manuscript.

Mala Bhattacharya: Concept and design.

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