

Neonatal bilateral ovarian cystadenoma: conservative or surgical treatment?

Cistoadenoma ovárico bilateral neonatal, ¿tratamiento conservador o quirúrgico?

Emilio Zenteno Salazar^a, Edgar Escalante Lucero^a, Carlos Valenzuela Dionicio^a, Orlando Miranda Gutiérrez^a, Arsenio Hernández Flota^{a,b}, Nina Méndez Domínguez^c, Juan C. Núñez Enríquez^d

^aUniversidad Marista de Mérida, Campus de Ciencias de la Salud. Mérida, México

^bHospitales Star Médica Mérida. Mérida, México

^cSubdirección de Enseñanza e Investigación. Hospital Regional de Alta Especialidad de La Península de Yucatán. Mérida, México

^dUnidad de Investigación Médica en Epidemiología Clínica, UMAE Hospital de Pediatría "Dr. Silvestre Frenk Freund" Centro Médico Nacional Siglo XXI, Instituto Mexicano del Seguro Social. Ciudad de México, México

Received: March 19, 2021; Approved: August 12, 2021

What do we know about the subject matter of this study?

The approach to ovarian cysts (OC) in the neonatal period is a clinical challenge. To our knowledge, there are no guidelines or consensus regarding the definitive management of OC in neonates.

What does this study contribute to what is already known?

We describe the case of a newborn with prenatal diagnosis of bilateral complex ovarian cysts, successfully operated laparoscopically, and discuss the conditions for conservative or surgical management according to the ultrasound characteristics of the cyst.

Abstract

Bilateral complex ovarian cysts in newborns are rare and their reporting becomes imperative to increase knowledge about the best therapeutic management. **Objective:** To describe the clinical case of a newborn with a diagnosis of bilateral complex ovarian cysts and to discuss the conditions for conservative or surgical management according to the ultrasound characteristics of the cyst. **Clinical Case:** At 35 weeks of gestational age, prenatal ultrasound identified the presence of cystic masses in both adnexa, so it was decided to interrupt the pregnancy by cesarean section at 37 weeks. After birth, bilateral ovarian cysts of 3.5 x 4.4 x 2.7 and 3.4 x 2.4 x 3.3 cm, right and left, respectively, were corroborated. The right cyst had a septum of 1.4 mm thick and thickened wall of 3 mm which was compatible with complex cysts. On the 4th day of extrauterine life, laparoscopic vacuum aspiration and deroofting with electrocautery of the upper wall of both cysts was performed, without complications. The diagnosis of ovarian serous cystadenoma was made by pathological anatomy. **Conclusions:** We describe a case with adequate prenatal diagnosis and laparoscopic surgical intervention of a bilateral

Keywords:

Neonatal Ovarian Cyst;
Cystadenoma;
Laparoscopy

ovarian cyst > 4 cm. Prenatal diagnosis is essential for choosing the best therapy management (conservative or surgical) depending on the echography characteristics of the cyst. Neonatal surgery is recommended for simple ovarian cysts > 4 cm, complex cysts regardless of their size, and those that become complex cysts during conservative management.

Introduction

Ovarian cysts (OC) represent the most frequent abdominal tumor in newborns, and it is possible to detect them prenatally in approximately 1 out of every 2500 pregnancies^{1,2,3}. They can be detected by ultrasound as early as the third trimester of gestation. They are considered pathological when they reach a diameter of 2 cm or more and can be confused with genitourinary, gastrointestinal cysts, or lymphangiomas^{4,5}.

The OC originates in the ovarian follicles and its main trigger for their development is the follicle-stimulating hormone (FSH) secreted by the fetal pituitary gland, followed by maternal estrogens and placental human chorionic gonadotropin (hCG). At birth, a significant decrease in the levels of FSH, circulating estrogens, and hCG occurs, which generally helps the OCs to involute and resolve spontaneously before one year of life^{6,7}. Likewise, it has been reported that ovarian tumors detected in the intrauterine stage are more frequently observed in fetuses of mothers with endocrinological disorders such as diabetes mellitus, gestational diabetes, and hypothyroidism⁴.

As described by Nussbaum in 1988⁸, ovarian cysts are classified by ultrasound according to their content in 2 types: simple cysts (SC), with anechoic, homogeneous content, larger than 2 cm in diameter, with a thin, barely perceptible wall, without solid components or fluid-fluid or fluid-debris level; and complex (CC), with thick wall (≥ 3 mm), presence of the double-wall sign (i.e., internal echogenic and external hypoechoic walls), hyperechoic content, fluid-fluid or fluid-debris level, and intracystic septa, the latter considered to be the result of ovarian torsion or intracystic bleeding^{9,10}.

OCs present variable evolution, most of them resolve spontaneously. Bascietto et al, state that the resolution of OCs is 54% overall and 80% in those < 4 cm in diameter^{1,8}. However, when the ultrasound characteristics are consistent with a complex ovarian cyst (CC), complications such as torsion and/or hemorrhage may occur, putting the ovarian tissue at risk after birth. This is the main complication of fetal-neonatal ovarian cysts, accounting for 30-78% of torsions¹¹. Most of them are asymptomatic, so those cysts that are not suitable for surgery should be followed by ultrasound, in order to observe the evolution from simple to complex cyst and thus suspect a pedicle torsion promptly¹¹.

The objective of this work is to describe the clinical case of a newborn with prenatal diagnosis of bilateral complex ovarian cysts, operated laparoscopically, as well as to discuss the conditions for conservative or surgical management according to the ultrasound characteristics of the cyst.

Clinical Case

Newborn female patient, first child of a 26-year-old mother, with adequate prenatal control and without pathological personal history relevant to the current picture. During gestation, a total of four fetal ultrasound studies were performed (weeks 14, 28, 32, and 35). In the prenatal ultrasound performed at 28 weeks of gestational age (WGA), cystic lesions at the renal level were observed for the first time, with differential diagnosis of renal cysts. She continued under medical supervision, however, it was not until the 35th week of gestation that two cystic images were identified, compatible with OC (figure 1); also, oligohydramnios was reported (4.7 amniotic fluid index), so it was decided to perform a cesarean section at 37 weeks of gestation.

The newborn was born weighing 2,580 grams, length 47 cm, head circumference 33 cm, with no perinatal complications. On physical examination, no malformations were observed, abdominal masses were palpated in both lower quadrants, so abdominal ultrasound was requested (figure 2). The ovarian cystic masses were identified at the level of both adnexa, the right one of 3.5 x 4.4 x 2.7 cm, 23 ml volume, 1.4 mm septum, and 3 mm thickened wall; and the left one of 3.4 x 2.4 x 3.3 cm, 14.7 ml volume, and 1 mm septum; with no increase in the vascular flow pattern in the lesions, nor free fluid in the abdominal cavity. The ultrasound diagnostic impression was of complex ovarian cysts.

On the fourth day of extrauterine life, a three-port laparoscopic exploration was performed under general anesthesia, visualizing in both iliac fossae dependent cystic lesions of both ovaries, with slight torsion (180° of torsion on its axis) of the right ovarian pedicle, without evidence of hypoperfusion or necrosis (figure 3A). Percutaneous evacuation of both cysts was performed via laparoscopy obtaining 20 and 10 ml of citrine-like fluid on the right and left side, respectively.

Subsequently, both cysts were unroofed, removing the upper wall of the cyst by electrosurgery, preserving the underlying ovarian tissue (figure 3B). Hemostasis was confirmed and the procedure was concluded without complications. The histopathological analysis identified a bilateral ovarian serous cystadenoma, with a smear negative for malignant cells.

There were no complications during her hospital stay, nor was there evidence in ultrasound imaging of lesions recurrence at 2 and 6 months of age.

Discussion

Since the advent of ultrasonography, prenatal diagnosis of ovarian lesions or cysts has improved but, despite this, the optimal management of fetal OCs is not standardized and many cases have been managed at the discretion of the specialized physician; however, there is a consensus on their management according to relevant ultrasound criteria to be considered in each case¹².

According to the classification of Bascietto et al, the case presented corresponds to a bilateral complex ovarian cyst, which has a torsion rate seven times higher than simple OC (44.9% vs. 6%, respectively)⁹. However, there is controversy and discussion about the diameter to consider (> 4 or > 5 cm) to decide surgical management. In this regard, it has been reported that OCs larger than 5 cm in diameter have a much higher

risk of complications, and 85% will require oophorectomy¹³. In addition, it has been shown that cysts smaller than 5 cm in size can also be associated with torsion¹⁴⁻¹⁷.

Pedicle torsion can be asymptomatic or present with pain, vomiting, fever, abdominal distension, leukocytosis, and peritonitis¹⁸. In the presence of pedicle torsion, emergency surgery is necessary to save the affected adnexa. In a study by Kim et al, 11 patients with torsion were asymptomatic, but in the postnatal follow-up ultrasound, complex cysts appeared with signs of internal bleeding such as echogenic debris inside and a fluid-fluid level, as well as a thickened wall¹⁴. Cysts > 4 cm have a higher risk of becoming complex in intrauterine life compared with those < 4 cm (OR = 3.16)⁹. Therefore, these ultrasound findings should be considered to suspect pedicle torsion in patients under conservative management¹⁴.

Regarding ovarian preservation, in a retrospective study that included 39 neonates with OC, of the three patients that required oophorectomy, the pathology sample reported viable ovarian tissue, therefore, it was recommended ovarian preserving surgery in all OC that undergo surgery for suspected ovarian torsion in the follow-up ultrasound¹¹. Considering the above, the case described had a low probability of spontaneous resolution and a high risk of pedicle torsion, due to its complex ultrasound characteristics (septa inside it and thickened wall ≥ 3 mm). The right adnexa was found with 180° torsion, without evidence of ischemia or ne-

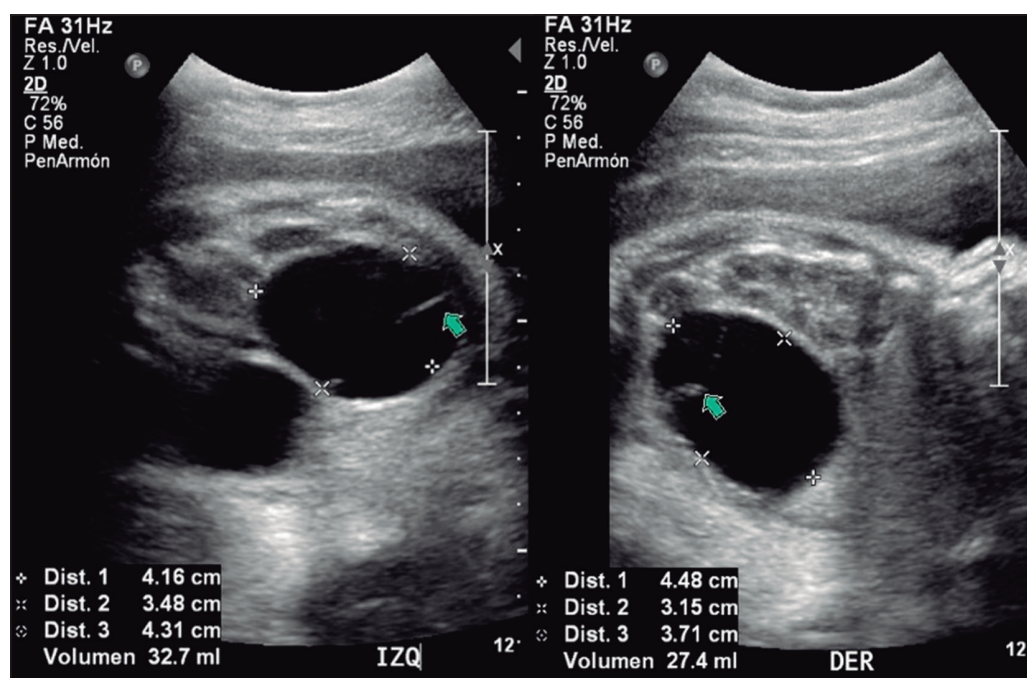


Figure 1. Prenatal control ultrasound (35 weeks gestation) with oligohydramnios (AFI 4.7) and cysts at the abdominopelvic level of 4.1 x 3.4 x 4.3 cm, 32 mL volume (left) and 4.4 x 3.1 x 3.7 mm, 27 mL volume (right) with septum inside (green arrow). AFI: amniotic fluid index.

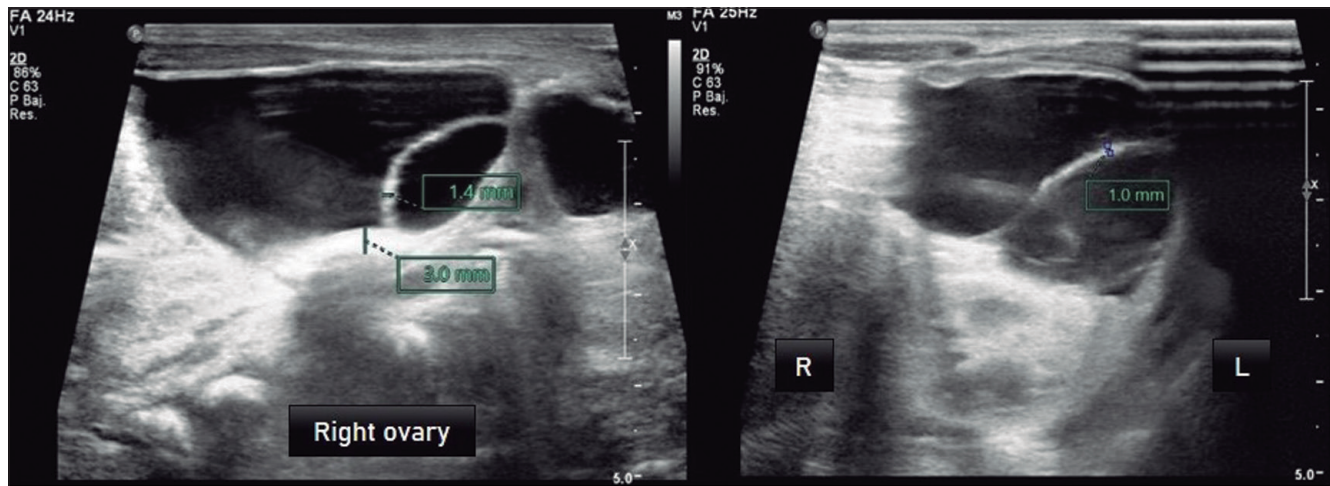


Figure 2. Postnatal ultrasound in the first 24 hours of extrauterine life. Ovarian cysts of 3.4x2.4x3.3 cm, 14.7 mL in volume with a 1 mm septum (left), and 3.5x4.4x2.7 cm, 23 mL with 1.4 mm septum (right).

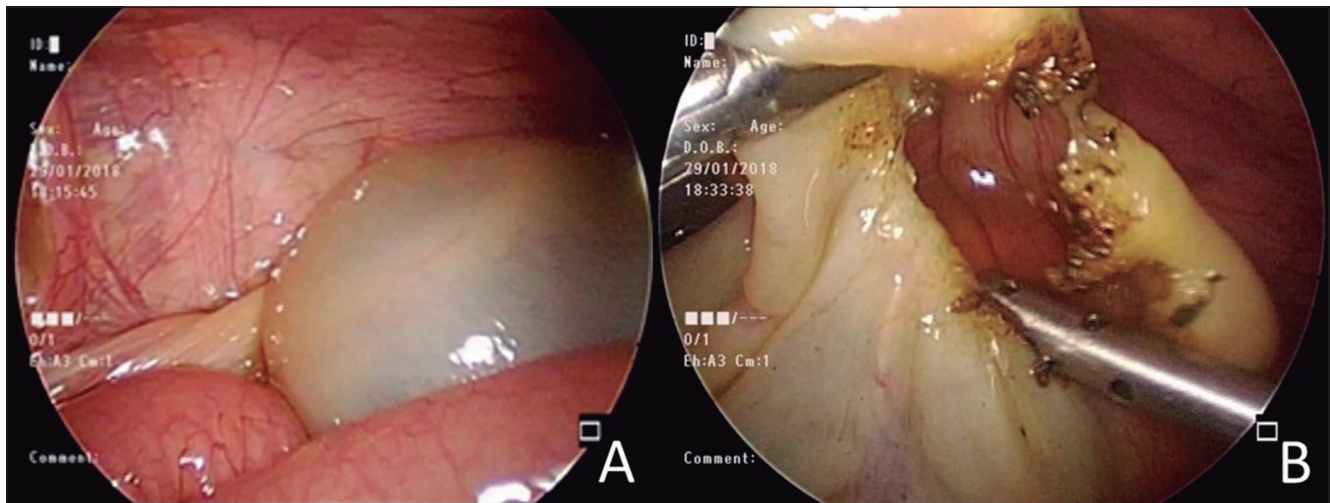


Figure 3. A) Laparoscopic view of the right ovary with cystic tumor and torsion. B) Laparoscopic view: Right ovarian cyst unroofed by electrocautery. The cystic fluid was amber color.

crisis, so unroofing of the upper wall of the cysts was performed.

Most of the literature favors conservative over surgical management for the resolution of simple OC < 4 cm^{4,7,9,19,20,21}. Conservative management consists of performing serial ultrasound scans every 2-3 weeks, however, there is a 20% risk of conversion of the OC into a complex one, increasing the risk of ovarian torsion and subsequent loss of the ovary^{9,22}. Therefore, although ultrasound findings are not established to define failure of conservative management, in the study by Papic et al, 10/11 (91%) of the patients under medical supervision who underwent surgery, an increase in size or a reduction of less than 10% of the cystic volume was observed, as well as conversion from simple to complex¹¹.

It has been suggested that surgical treatment of neonatal ovarian cysts should be indicated when it is a simple cyst > 4 cm, a complex cyst regardless of size, a cyst with progressive increase in size that persists beyond 6-12 months of age, or that present symptoms^{4,20,21}. The classic surgical approach has been infraumbilical laparotomy; however, the laparoscopic approach which is a minimally invasive procedure has been the preferred one in recent years^{17,21} because it offers advantages over laparotomy such as that it is easy to access, offers security at the moment of panoramic visualization, which allows confirming the diagnosis in case of doubt by visualization and biopsy; it reduces postoperative pain, presents the lowest incidence of postoperative adhesions, and is well tolerated by neonates^{1,21,23}.

Another group of pediatric surgeons has had good results and recommends neonatal aspiration guided by ultrasound or laparoscopy for simple cysts²⁴. In a study by Cho et al, which included 14 patients who underwent aspiration, no cystic evidence was observed on follow-up ultrasound in 11/14 patients and in 3/14 patients a cyst < 2 cm was observed. They also recommend laparoscopic-guided aspiration in OC > 4 cm or complex cysts without torsion²⁴.

In recent years, the possibility of *in utero* aspiration (IUA) has been studied to reduce the risk of prenatal ovarian torsion, however, its role is still controversial. In a multicenter, prospective, randomized study of 66 pregnant mothers of fetuses over 28 WGA with OC > 3 cm without evidence of complexity, were divided into two groups as expectant management group (n = 27) and intrauterine aspiration group (n = 34)²⁵. IUA was associated with a higher incidence of *in utero* cyst involution (47.1% vs 18.5%; RR, 2.54) and lower incidence of oophorectomy (3.0% vs 22.2%; RR, 0.13), however, it was not associated with a reduction in the need for surgical intervention in the first days of extrauterine life (37% vs 20.6%, respectively)²⁴. Noia, et al (2011) recommend IUA in OC > 3.5 cm, in order to decrease the risk of prenatal ovarian torsion, despite that, in their study, 76.9% of aspirated cysts recurred prenatally²⁶, while Bascietto et al, in their systematic review, mention that 37.9% had intrauterine recurrence⁹. IUA could be considered especially in the case of large cysts that present before birth, in order not to compromise the reproductive capacity of the fetus in adult life or in case the cyst causes compression in adjacent structures, which leads to obstruction or alteration of the flow in the venous duct⁹.

In this case, considering the bilaterality, size (> 4 cm), and ultrasound features of a complex cyst, it was decided to perform laparoscopy for resection of both OCs by percutaneous evacuation and unroofing of the upper wall of the cyst, in order to preserve the underlying healthy ovarian tissue. Since prenatal diagnosis was performed promptly, preservation of both ovaries was possible, and the patient's prognosis was favorable.

Conclusion

The approach to ovarian cysts in the neonatal pe-

riod is a clinical challenge. To our knowledge, there are no guidelines or consensus regarding the definitive management of OCs in neonates, however, there are recommendations from experts or based on review studies where it has been reported that it has been possible to resolve OCs successfully in most cases. Prenatal diagnosis in the third trimester of gestation is essential for a timely conservative or surgical approach, always considering the ultrasound characteristics of the lesion. It is recommended that simple cysts < 4 cm be managed conservatively, while minimally invasive surgical management (unroofing or laparoscopic-guided aspiration) will be chosen in those simple cysts > 4 cm or with complex ultrasound features. Although IUA has an impact on the reduction of pedicle torsion, it needs more studies to support it and to be performed by specialized health professionals.

Ethical Responsibilities

Human Beings and animals protection: Disclosure the authors state that the procedures were followed according to the Declaration of Helsinki and the World Medical Association regarding human experimentation developed for the medical community.

Data confidentiality: The authors state that they have followed the protocols of their Center and Local regulations on the publication of patient data.

Rights to privacy and informed consent: The authors have obtained the informed consent of the patients and/or subjects referred to in the article. This document is in the possession of the correspondence author.

Conflicts of Interest

Authors declare no conflict of interest regarding the present study.

Financial Disclosure

Authors state that no economic support has been associated with the present study.

References

1. Cabellos M, Cavieres C, Zarges T, Gajardo C, Vega D. Quiste ovárico fetal: diagnóstico prenatal y manejo quirúrgico postnatal exitoso. *Rev Chil Obstet Ginecol.* 2012;77(4): 306-9.
2. Youssef AA, Marei MM, Abouelfadl MH, Mahmoud WM, Elbarawy AS. Destechado de quistes extracorpóreo transumbilical asistido por laparoscopia para el tratamiento de quistes ováricos neonatales. *Egipto J Surg* 2020; 39: 177-82.
3. Valenti C, Kassner EG, Yermakov V, Cromb A. Antenatal diagnosis of a fetal ovarian cyst. *Am J Obstet Gynecol.* 1975;5:216-9.
4. Tyraskis A, Bakalis S, David A, Eaton S, De Coppi P. A systematic review and meta-analysis on fetal ovarian cysts: impact of size, appearance and prenatal aspiration. *Prenat Diagn.* 2017;37(10):951-8.
5. Trinh T, Kennedy A. Fetal Ovarian Cysts: Review of Imaging Spectrum, Differential Diagnosis, Management, and Outcome. *RadioGraphics.* 2015;35(2):621-35.
6. Noia G, Riccardi M, Visconti D, et al. Invasive fetal therapies: Approach and results in treating fetal ovarian cysts. *J Matern Fetal Neonatal Med.* 2012; 25:299-303.
7. Dolgin, S. Ovarian Masses in the Newborn. *Seminars in Pediatric Surgery.* 2000;9(3):121-7.
8. Nussbaum A, Sanders R, Hartman D, Dudgeon D, Parmley T. Neonatal ovarian cysts: sonographic-pathologic correlation. *Radiology.* 1988;168(3):817-21.
9. Bascietto F, Liberati, M, Marrone L, et al. Outcome of fetal ovarian cysts diagnosed on prenatal ultrasound examination: systematic review and meta-analysis. *Ultrasound Obstet Gynecol.* 2017;50:20-31.
10. Prasad S, Chui C. Laparoscopic-Assisted Transumbilical Ovarian Cystectomy in a Neonate. *JSLS.* 2007; 11(1), 138-41.
11. Papic J, Billmire D, Rescorla F, Finnell S, Leys C. Management of Neonatal Ovarian Cysts and its Effect on Ovarian Preservation. *J Pediatr Surg.* 2014;49(6):990-4.
12. Camoglio F, Bianchi F, Peretti M, et al. Management of Neonatal Ovarian Cysts: Clinical Aspects. *Int J Gynecol Clin Pract.* 2017; 4: 130-2.
13. Bagolan P, Giorlandino C, Nahom A, et al. The management of fetal ovarian cysts. *J Pediatr Surg.* 2002;37(1):25-30.
14. Kim H, Yoo S, Cha M, Kim J, Jeon T, Kim W. Diagnosis of neonatal ovarian torsion: Emphasis on prenatal and postnatal sonographic findings. *J Clin Ultrasound.* 2016;44(5):290-7.
15. Signorelli M, Gregorini M, Platto C. The prognostic value of antenatal ultrasound in cases complicated by fetal ovarian cysts. *J Neonatal Perinatal Med.* 2019;12(3):339-43.
16. Grapin C, Montagne JP, Sirinelli D, Silbermann B, Gruner M, Faure C. Diagnosis of ovarian cysts in the perinatal period and therapeutic implications (20 cases). *Ann Radiol (Paris).* 1987;30(7):497-502.
17. Youssef A, Marei M, Abouelfadl M, Mahmoud W, Elbarawy A. Laparoscopy-assisted transumbilical extracorporeal cyst deroofing for the management of neonatal ovarian cysts. *Egypt J Surg.* 2020; 39:177-82.
18. Fuentealba T. Quistes ováricos en recién nacidas, niñas y adolescentes: aspectos ultrasonográficos. *Rev Chil Radiol.* 2006;12(1):15-20.
19. Tajiri T, Souza R, Kinoshita Y, et al. Surgical intervention strategies for pediatric ovarian tumors: experience with 60 cases at one institution. *Pediatr Surg Int.* 2011;28(1):27-31.
20. Manjiri S, Padmalatha SK, Shetty J. Management of Complex Ovarian Cysts in Newborns-Our Experience. *J Neonatal Surg.* 2017;6:3.
21. Hashish A. Minimally invasive surgery for ovarian cysts in children. *Annals of Pediatric Surgery.* 2011; 7(4):117-22.
22. Álvarez A, Santomil P, Serna J, Chávez L, Gallart R, Martínez E. Tratamiento quirúrgico de los quistes ováricos perinatales. *Cir Pediatr.* 2010;23: 225-8.
23. Corbett J, Lamont A. Bilateral ovarian autoamputation in an infant. *J Ped Surg.* 2002;37(9):1359-60.
24. Cho M, Kim D, Kim S. Ovarian Cyst Aspiration in the Neonate: Minimally Invasive Surgery. *J Pediatr Adolesc Gynecol.* 2015;28(5):348-53.
25. Diguisto C, Winer N, Benoist G, et al. In-utero aspiration vs expectant management of anechoic fetal ovarian cysts: open randomized controlled trial. *Ultrasound Obstet Gynecol.* 2018; 52(2):159-64.
26. Noia G, Riccardi M, Visconti D, et al. Invasive fetal therapies: Approach and results in treating fetal ovarian cysts. *J Matern Fetal Neonatal Med.* 2012; 25:299-303.