Pak. j. life soc. Sci. (2024), 22(2):10795-10801 E-ISSN: 2221-7630;P-ISSN: 1727-4915

Pakistan Journal of Life and Social Sciences
www.pjlss.edu.pk

https://doi.org/10.57239/PJLSS-2024-22.2.00816

RESEARCH ARTICLE

Large Paratubal Cyst in Pregnancy with ASD Secundum L to R Shunt: A Rare Case Report

Huwaida Nabilah[1*], Nareswari Imanadha Cininta Marcianora[2], Birama Robby Indraprasta[3]

[1]Obstetric and Gynecology Resident of Airlangga University, Dr. Soetomo General Academic Hospital, Surabaya, Indonesia

[2]Staff of the Fetomaternal Division, Obstetric and Gynecology Department, Airlangga University, Dr. Soetomo General Academic Hospital, Surabaya, Indonesia

[3]Staff of the Oncology and Gynecology Division, Obstetric and Gynecology Department, Airlangga University, Dr. Soetomo General Academic Hospital, Surabaya, Indonesia

ARTICLE INFO

Received: Aug 12, 2024

Accepted: Sep 17, 2024

Keywords

Paratubal Cyst

Para-Ovarian Cyst

Ovarian Cyst

Paratubal Cyst In Pregnancy

Adnexal Mass In Pregnancy

***Corresponding Author:**

nabilahuwaida90@gmail.com

ABSTRACT

Paratubal cysts account for 5–20% of adnexal masses. It develops from the mesosalpinx or broad ligament, located near the ovary and Fallopian tube. Large cyst may impact fetal growth, causing malpresentation, obstructed birth, ruptured cyst, inaccurate gestational age estimate, and increase maternal morbidity. Objective: To describe a case of a large paratubal cyst in pregnancy. A 39/40 weeks pregnant 30-years-old women with a known ovarium cyst since 2019 was referred with large atrial septal defect (ASD), dominant R-to-L shunt, mild tricuspid regurgitation, and probable severe pulmonary hypertension. On admission, biometry matched the pregnancy of 35/36 weeks gestational age. We observed a cyst sized 6.79 x 5.64 x 12.01 cm in the right adnexa with intrauterine growth restriction of the fetus. Echocardiographic showed secondary ASD to L-to-R shunt and intermediate pulmonary hypertension. Intraoperative finding includes 17x12 cm right paratubal cyst, containing 1000 cc of brown fluid during excision. Caesarean section, IUD implantation, and right paratubal cyst excision were performed. Histopathological evaluation suggested a benign cyst with no sign of malignancy. Ovarian or para-ovarian/paratubal tumors during pregnancy are uncommon. Ultrasound is highly sensitive and specific in detecting these masses. Cysts greater than 20 cm in diameter may cause problems related to increased pressure on the lower abdominal cavity. Furthermore, there is a higher risk of torsion, which causes severe pain. Tumors are usually discovered incidentally during a routine examination, ultrasound, or during cesarean section. Surgical treatment has been recommended for ovarian masses larger than 6 cm in diameter if symptomatic. Both diagnosis and management for paratubal cysts during pregnancy are challenging therefore. The most reliable diagnostic method for confirming gynaecological diagnoses is ultrasonography. Treatment options vary depending on maternal age and parity, also the size of the cyst.

INTRODUCTION

Adnexal masses are one of the most common pregnancy complications, with an incidence of 1- 5.3% (1). The majority of adnexal masses during pregnancy are functional or benign, with only about 5% being malignant (Martone et al., 2021). However, in women, malignancy is the second most common cause of death (Brahmantara et al., 2020). Paraovarian and paratubal cysts are a common cause of adnexal masses (Barloon et al., 1996). In pathologically verified series, paratubal cysts account for 5 - 20% of all adnexal masses (Savelli et al., 2006). They are typically asymptomatic and

benign, with a reported incidence of malignancy of 2-3% (Savelli et al., 2006). The majority of paratubal cysts are misdiagnosed as ovarian cysts, and only one out of every fifteen patients is suspected before surgery (Tjokroprawiro, 2021). Histopathologically, they are divided into three groups: paramesonephric, mesonephric, and mesothelial (Savelli et al., 2006). A paratubal cyst develops from the mesosalpinx or broad ligament, which is located near the ovary and the Fallopian tube (Savelli et al., 2006). Large size can impact foetal growth, malpresentation, obstructed birth, rupture of the cyst, incorrect gestational age estimate, and consequently increase maternal morbidity from excessive abdominal distension (Hota et al., 2015).

Case Illustration

A 30-year-old woman with 39/40 weeks of pregnancy has been identified as having an ovarium cyst since 2019. She was scheduled for surgery. A cardiac disorder was discovered during the pre-operation evaluation, so the surgery was canceled. The cardiac disorder was not further diagnosed. The patient did not take any medication and had no heart problems. At 24/25 weeks of pregnancy, she had an echocardiographic examination at Dr. R. Koesma Tuban's General Regional Hospital, which revealed a large atrial septal defect (ASD) and mild pulmonary hypertension. The patient was advised to have regular check-ups at the Maternal Unit of the General Regional Hospital of Tuban. She did not, however, perform any medical checks at the cardiac unit. She was then re-evaluated during the surgical preparation at 38/39 weeks of pregnancy. The echo result revealed a large ASD with a dominant R-to-L shunt, mild TR, and a high probability of severe PH. The patient was then referred to Dr. Soetomo Hospital's Maternal Unit.

When examined at Dr. Soetomo Hospital's Maternal Unit, the patient's biometry matched the pregnancy age of 35/36 weeks and EFW 2534 g (p 4.1%). A cyst measuring 6.79 x 5.64 x 12.01 cm in the right adnexa was discovered during the USG examination (Figure 1), as well as intrauterine growth restriction (IUGR) in the fetus. Dr. Soetomo Hospital's echocardiographic result is an ASD Secundum L to R shunt with an intermediate probability of PHT. Discussions with Dr. Soetomo Hospital's cardiac expert team yielded some results. The patient had an ASD left to right shunt as well as an intermediate probability of PHT. She had mWHO class IV as a result of PAH. She was diagnosed with GI P0000 39/40 wga, singleton pregnancy, ASD Secundum L to R shunt, intermediate probability of PHT, right ovarian cyst, and IUGR in the fetus. The patient was scheduled for a Caesarean section as well as a cystectomy.

Findings during the operation showed the gravid uterus, left ovary, and fallopian tube within the normal limit. The findings also showed that the right paratubal cyst was 17 cm by 12 cm (Figure 2). During the excision, the cyst popped and produced brown fluid, approximately 1000 cc. Thus, the patient was then assigned for a Caesaran section with IUD insertion and excision in the right paratubal cyst. The baby weighs 2,500 g with a 50 cm height, an Apgar score of 8–9, a Ballard score of 40 weeks, and a Lubchenco score of p < 10. The patient's histopathological results show no evidence of malignancy. Paratubal cysts containing benign cysts are present.

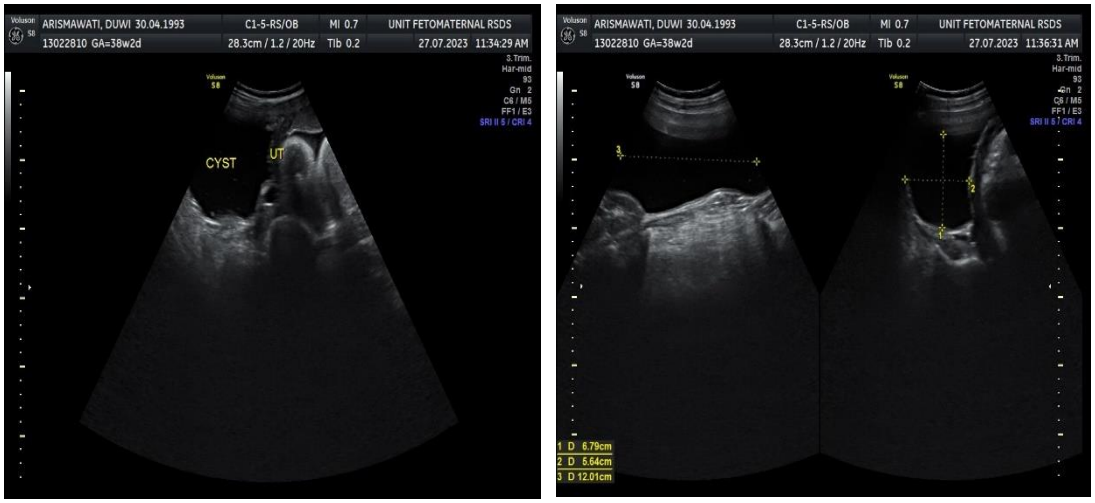

Figure 1: The USG evaluation showed a cyst measuring 6.79 x 5.64 x 12.01 cm in the right adnexa.

Figure 2: Images showing the results of surgery. The right paratubal cyst measured 17 cm by 12 cm. The right ovary, left ovary, and left fallopian tube are all within normal limits.

DISCUSSION

The paratubal cyst, also known as the paraovarian cyst, is a foetal anatomical structure that is thought to have originated from the Wolffian duct (Thanasa et al., 2023). It was initially identified as a distinct clinical entity in 1973 by Kariminejad and Scully (Thanasa et al., 2023). The terms "paratubal" and "paraovarian" are used in proportion to how far the cyst is from the ovary or fallopian tube, respectively (Thanasa et al., 2023). It is estimated that the mesothelium of the broad ligament is the source of the condition in about two-thirds of instances, whereas the paramesonephric or mesonephric remnants are the source in 30% and 2% of cases, respectively (Thanasa et al., 2023). Typically, they are no larger than 75 mm, but in 13% of uncommon instances, the cyst's maximal diameter might be greater than 150 mm (Thanasa et al., 2023). Paraovarian or paratubal cysts typically manifest in the third or fourth decade of life and comprise 5% to 20% of all adnexal masses (Thanasa et al., 2023). Preadolescence is when it happens less frequently, and in a very rare instance (1/1500000), it can result in isolated torsion of the ipsilateral fallopian tube (Thanasa et al., 2023). Bland-Sutton (1890) was the first to report isolated torsion of the fallopian tube around its longitudinal axis, which is an uncommon cause of acute abdomen (Thanasa et al., 2023). Malignant transformation is also incredibly rare (Thanasa et al., 2023). It is believed that when a paraovarian or paratubal cyst measures more than 5 cm, the likelihood of cancer increases (Thanasa et al., 2023).

Ovarian or para-ovarian/paratubal tumors during pregnancy are rare (Kumbak et al., 2010). Paratubal cysts are usually small and asymptomatic (Thanasa et al., 2023). Large lesions, which are cysts greater than 20 cm in diameter, cause problems. These issues arise due to pressure on the lower abdominal cavity. Additionally, torsion of the paratubal cyst can cause complications. These large cysts are associated with mild, dull, low-intensity stomach discomfort. They can also lead to the rapid onset of acute abdominal pain (Thanasa et al., 2023).

In the case of this patient, the paratubal cyst did not cause symptoms (Thanasa et al., 2023). This is because its size was approximately 17 cm by 12 cm. The size being less than 20 cm prevented symptom manifestation. Typically, larger cysts would present with noticeable discomfort. However, in this patient, no such symptoms were observed. The absence of symptoms is directly linked to the cyst's size. Larger cysts often lead to significant pressure-related issues. They also have a higher risk of torsion, which causes severe pain (Thanasa et al., 2023).

In this scenario, the patient's cyst remained under the threshold that typically causes complications. Therefore, no immediate discomfort was reported. The cyst's size played a crucial role in the patient's symptomatology. Had the cyst been larger, different symptoms would likely have been present.

Fortunately, this patient did not experience those issues. The cyst's dimensions were a determining factor in the absence of symptoms. Consequently, the patient's condition was more manageable (Thanasa et al., 2023).

In summary, the size of paratubal cysts is critical in symptom development. This patient's cyst size was below the threshold for causing notable symptoms (Thanasa et al., 2023).

It might be difficult to diagnose a large abdominal and pelvic mass clinically, especially if it occurs concurrently with pregnancy (Santos et al., 2015). The tumors are usually discovered by chance during a routine examination, ultrasound, or a cesarean section at term (Kumbak et al., 2010). The most reliable diagnostic method for confirming gynaecological diagnoses is ultrasonography, which has a 92% sensitivity and 96% specificity (Indirayani et al., 2023). A round or oval cyst that is next to the ipsilateral ovary but distinct from it can be found using ultrasound (Alpendre et al., 2020). The diagnosis is further aided by the split sign, which is the cyst's separation from the ovary during an endovaginal probe movement (Alpendre et al., 2020). However, particularly if these cysts are large, diagnosis becomes challenging when they are located in close proximity to the ovary or tube (Alpendre et al., 2020). Differential diagnosis by ultrasound includes ovarian cyst, hydrosalpinx and peritoneal inclusion cyst (Alpendre et al., 2020). However, multiloculated, solid tumours with thick septae containing nodularity, papillary projections, or Doppler flow-indicating neovascularization are ultrasonographic characteristics that raise greater concerns regarding malignancy (Archer et al., 2023). Paratubal cysts have low signal intensity on MRI with T1WI. If significant signal intensity is seen inside the cyst, this may indicate haemorrhage from torsion, hence careful examination is necessary (Kim et al., 2023). On T2WI, however, paratubal cysts have a strong signal, and internal heterogeneity in the cysts points to torsion-related haemorrhage (Kim et al., 2023). Moreover, CT scanning can reveal lymph nodes or the omentum as sites of metastasis; yet, sonography is usually better when the disease is restricted to the pelvis (Archer et al., 2023). In this case, ultrasound discovered a cyst measuring 6.79 x 5.64 x 12.01 cm in the right adnexa.

Paraovarian cysts are typically unilocular. They are filled with clear fluid. These cysts originate in the paramesonephric, mesothelial, or mesonephric regions (Qian et al., 2021). Histopathological examination shows specific cellular characteristics. Cells from the paramesonephric origin are secretory and ciliated. These cells also include low cuboidal epithelium. Occasionally, there are transparent cells from the mesonephric origin (Qian et al., 2021). The fibrous tissue around these cysts has a mesothelial origin. This tissue is surrounded by flattened epithelium. Sometimes, this epithelium shows tubal differentiation (Qian et al., 2021). However, the epithelium is often disrupted. This disruption is due to the distention of the cyst cavity. Therefore, it is difficult to make an absolute distinction between cell types.

In the case presented, histopathological anatomy results were examined. These results showed no signs of malignancy. The paratubal cysts were found to contain benign cysts. This confirms the absence of any malignant features (Qian et al., 2021). The presence of benign cysts was evident. There were no indications of cancerous changes. The cellular characteristics supported a benign diagnosis. The fibrous tissue showed typical mesothelial origin features. The cysts maintained their unilocular structure. The clear fluid content was consistent with paraovarian cysts. Overall, the findings indicated a benign condition (Qian et al., 2021).

Torsion or rupture of the mass may occur occasionally during pregnancy (Kumbak et al., 2010). Guidelines for the treatment of fallopian tube torsion have not yet been established (Syed et al., 2021). However, because it presents as acute abdominal or pelvic discomfort, diagnostic laparoscopy or exploratory laparotomy are typically used to make the final diagnosis (Syed et al., 2021). When a patient is hemodynamically stable and there is access to expertise, laparoscopy, the gold standard, is preferred (Syed et al., 2021). The cyst's size is also important; in accordance with the Royal College of Obstetricians and Gynaecologists' (RCOG) guidelines on the management of ovarian cysts in premenopausal women, cysts larger than 7 cm are more likely to rupture during surgery (Syed et al., 2021). Less post-operative pain, feverish morbidity, a quicker recovery, lower costs, and an early hospital discharge are among the advantages of laparoscopic surgery (Syed et al., 2021). The impact of tubal function and fertility following detorsion is not well-established (Syed et al., 2021). Given the information at hand, detorsion is advised in cases without gangrenous changes and salpingectomy is

advised in cases with delayed gangrenous changes (Syed et al., 2021). Diagnosis is challenging because signs and symptoms are not often specific enough to alert the doctor to the likelihood of fallopian tube torsion secondary to paraovarian or paratubal cyst torsion (Syed et al., 2021). In this case, the patient was diagnosed with an ovarium cyst during an aterm pregnancy. However, the patient's paratubal cyst diagnosis was made only during surgery. The characteristic of a paratubal cyst is a closed sac filled with fluid that develops in the wide ligament between the fallopian tube and the ovary (Farì et al., 2021). In this patient, the cyst ruptured during the excision, releasing about 1000 cc of dark fluid.

Furthermore, some adnexal masses are discovered by chance during a cesarean section (Kumbak et al., 2010). Treatment options vary depending on many factors, including age, parity, and size of the cyst (Atileh et al., 2020). Surgical management has been recommended for ovarian masses larger than 6 cm in diameter during the symptomatic period and in the second trimester to avoid miscarriage, torsion, rupture, and delayed diagnosis of malignancy (Kumbak et al., 2010). Moreover, the dependency of pregnancy on the corpus luteum for progesterone indicates that organogenesis has essentially finished (Nelson & MacKenzie, 2022). Laparotomies and laparoscopies are two methods of performing surgery (Permatasari & Sunur, 2024). Selecting the best surgical technique for excising an adnexal tumour while pregnant might be challenging (Permatasari & Sunur, 2024). The gold standard for treating benign adnexal masses is laparoscopy (Bhansakarya & Subedi, 2020). However, many surgeons choose laparotomy to laparoscopy when the cyst is large because of the increased risk of malignancy and the limited amount of room (Bhansakarya & Subedi, 2020). Laparoscopic approach for large cysts can only be attempted if the diagnosis is certain (Mohamed Farès et al., 2023). Adnexal mass excision via laparoscopy was associated with a decreased incidence of LBW, even though laparoscopy and laparotomy appear to be similarly equivalent, useful, and safe in terms of pregnancy outcomes (Permatasari & Sunur, 2024). When compared to laparoscopy, pregnant women who underwent open ovarian resection had a considerably higher risk of preterm birth and low birth weight (Permatasari & Sunur, 2024). A recent registry study conducted in Japan found that laparoscopic surgery improved short-term foetal adverse events, incidence of blood transfusions, operative time, and length of hospital stay (Permatasari & Sunur, 2024). In order to distinguish paratubal cysts from ovarian cysts, it is necessary to identify during surgery that blood vessels are passing over the cyst (Almahmeed et al., 2022). The decision was made to perform a paratubal cyst excision procedure on this patient. The cyst was +/- 17 cm x 12 cm in size and was performed concurrently with the caesarean section. With the histopathological anatomy results showing no signs of malignancy, the patient's planned therapy is follow-up care.

CONCLUSION

The diagnosis of a paratubal cyst during pregnancy is challenging, as is the procedure. The accuracy of the diagnosis has a significant impact on the procedures used to treat patients.

Acknowledgments

The authors would like to thank the patient for her willingness to share her medical history and participate in this case report. We would also like to acknowledge the contributions of the medical staff involved in the patient's care. Thank you to Obstetric and Gynecology Department, Faculty of Medicine, Airlangga University, Dr. Soetomo Hospital, Surabaya

Conflict of interest

The authors have no conflicts of interest.

Funding

This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

Ethical clearance

Ethical clearance was not required for this case report as it does not involve any experimental procedures or interventions. The patient's identity has been anonymized to protect her privacy.

Author contributors

Each author wrote the text and took part in the study's design. The final manuscript was read and approved by all writers. HN: manuscript writing. Gathering and managing data. NICM: developing protocols and projects, gathering and managing data. BRI: revision of the work, study of the literature, ideation and verification of manuscripts.

REFERENCES

Almahmeed E, A Alshaibani, H Alhamad and A Abualsel, 2022. Giant Paratubal Cyst Mimicking Mesenteric Cyst. Case Rep Surg, 2022: 4909614.

Alpendre F, I Pedrosa, R Silva, S Batista and P Tapadinhas, 2020. Giant paratubal cyst presenting as adnexal torsion: A case report. Case Rep Womens Health, 27: e00222.

Archer S, VI Alaniz and PS Huguelet, 2023. Surgical management of a giant paratubal cyst: a case report and review of the literature. Annals of Pediatric Surgery, 19: 15.

Atileh LIA, D Dahbour, H Hammo and M Abdullattif, 2020. Laparoscopic Removal of a 40-cm Paratubal Cyst in a Morbidly Obese Patient. Gynecol Minim Invasive Ther, 9: 39-41.

Barloon TJ, BP Brown, MM Abu-Yousef and NG Warnock, 1996. Paraovarian and paratubal cysts: preoperative diagnosis using transabdominal and transvaginal sonography. Journal of Clinical Ultrasound, 24: 117-122.

Bhansakarya R and S Subedi, 2020. Laparoscopic Management of Large Right Paratubal Cyst: A Case Report. JNMA: Journal of the Nepal Medical Association, 58: 501-504.

Brahmantara B, I Kartha and I Budiana, 2020. Characteristic gynaecology-oncology patients from Obstetrics and Gynaecology Outpatient Clinic Mangusada Regional Hospital Badung reffered to Sanglah Hospital from 1st January 2016 – 31st December 2018. Bali Medical Journal, 9: 430.

Farì G, M Dell'Aquila, S Moresi, A Lanzone and V Arena, 2021. Ectopic deciduosis in a paratubal cyst: A report of the first case. International Journal of Gynaecology and Obstetrics, 154: 376-378.

Hota BM, KR Rukmini and PGC Basavaih, 2015. Giant Ovarian cyst in Term Pregnancy-A rare case report. Bali Medical Journal, 4: 5-7.

Indirayani I, H Nora, CR Maharani, R Jannah, DMA Febrina and IC Mulya, 2023. A successful pregnancy complicated with ovarian cyst torsion in the first trimester. Bali Medical Journal, 12: 2176-2179.

Kim J, J Lim, JW Sohn, SM Lee and M Lee, 2023. Diagnostic imaging of adnexal masses in pregnancy. Obstet Gynecol Sci, 66: 133-148.

Kumbak B, H Celik, B Cobanoglu and B Gurates, 2010. Paratubal borderline tumor incidentally found during cesarean section: case report and review of literature. European Journal of Surgical Oncology, 36: 789-791.

Martone S, L Troìa and S Luisi, 2021. Adnexal masses during pregnancy: management for a better approach. Gynecological Surgery, 18: 3.

Mohamed Farès M, K Anis, BD Anis, E Nada, R Bochra, K Yasser and BM Mounir, 2023. Abdominal mass revealing a giant paratubal cyst. Archives of Gynecology and Obstetrics, 308: 1395-1396.

Nelson LW and E MacKenzie, 2022. Paratubal Cystectomy in a Pregnant Woman Using the Single-Incision Laparoscopic Surgery (SILS) Technique. Case Rep Obstet Gynecol, 2022: 2802767.

Permatasari P and YR Sunur, 2024. PARATUBAL TORSION CYST WITH NORMAL OVARY IN THE FIRST TRIMESTER OF PREGNANCY: A CASE REPORT. Teiko Medical Journal, 47

Qian L, X Wang, D Li, S Li and J Ding, 2021. Isolated fallopian tube torsion with paraovarian cysts: a case report and literature review. BMC Women's Health, 21: 345.

Santos VM, LA Santos and FR Modesto, 2015. Giant ovarian cyst. Bali Medical Journal, 4

Savelli L, T Ghi, P De Iaco, M Ceccaroni, S Venturoli and B Cacciatore, 2006. Paraovarian/paratubal cysts: comparison of transvaginal sonographic and pathological findings to establish diagnostic criteria. Ultrasound in Obstetrics and Gynecology, 28: 330-334.

Syed S, A Amin and M Ullah, 2021. Fallopian Tube Torsion Secondary to Paraovarian Fimbrial Cyst: A Difficult to Diagnose and a Rare Cause of Acute Abdomen in Adolescent. Cureus, 13: e17888.

Thanasa A, E Thanasa, EM Xydias, E Kamaretsos, I Paraoulakis, AC Ziogas, V Grapsidi, G Kontogeorgis, EE Gerokostas and I Thanasas, 2023. Pueperal Paratubal Cyst Torsion and Secondary Fallopian Tube Torsion Without Ovarian Involvement: A Case Report. Cureus, 15: e36540.

Tjokroprawiro BA, 2021. Huge Paratubal Cyst: A Case Report and a Literature Review. Clinical Medicine Insights: Case Reports, 14: 11795476211037549.