International Journal of Reproduction, Contraception, Obstetrics and Gynecology
Gadre S et al. Int J Reprod Contracept Obstet Gynecol. 2024 Sep;13(9):2532-2536
www.ijrcog.org
pISSN 2320-1770 | eISSN 2320-1789

DOI: https://dx.doi.org/10.18203/2320-1770.ijrcog20242517

Case Report

A rare case of bilateral paraovarian cysts, giant on one side

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Received: 26 June 2024
Revised: 21 July 2024
Accepted: 05 August 2024

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ABSTRACT

Paraovarian cyst is a type of adnexal cyst originating from the mesothelium in the broad ligament between the fallopian tube and the ovary; it accounts for 10% of all adnexal masses. They are considered giant when the threshold of 150 mm is exceeded. Only a few cases of giant paraovarian cysts (>20 cm) have been published, and all cases have had different approaches and histopathological types. The aim is to report a case of Bilateral paraovarian cyst in which one side cyst was giant, because of rarity to add to the literature. A 26-year-old nulligravida presented with mass per abdomen with no other associated complaints. She was taken for Exploratory laparotomy with subsequent bilateral paraovarian cyst aspiration with cyst excision. Frozen section sent to histopathology revealed benign serous cystadenoma. It draws clinical attention in the event of complications like cyst enlargement, torsion, rupture, haemorrhage and neoplasm. Age, gross appearance, size, septation, and Ca 125 levels are all weak indicators of malignancy. Hence, frozen sections should be checked intraoperatively to confirm the malignancy status, especially when there is a papillary projection. As a huge paraovarian cyst may mimic a large ovarian cyst hence it should be included in the differential diagnosis of pelvic masses, especially in the reproductive age.

Keywords: Paraovarian cyst, Case report, Giant on one side

INTRODUCTION

Lump per abdomen is a symptom which can have myriads of differential diagnoses in obstetrics and gynaecology. Unbeknown of its benign or malignant nature a giant mass per abdomen stipulates the need of a discrete diagnosis to pre-empt the further management of such patient. Paraovarian cysts (POCs) are cystic tumors that can be found between the ovarian hilum and the ovarian fimbria.1 It is located within the mesosalpinx and broad ligament and is usually diagnosed in the age of 30 to 40 years affirming higher incidence in reproductive age group.[1,2] Although they have been reported in all age groups from premenarchial period up to menopause.[3] They account for 10% of all adnexal masses. They are mostly unilateral, benign and measure between 0.2 cm to 2 cm in size with only few cases reported to exceed 15cm in diameter.[4] Traditionally, adnexal cysts are termed 'large' when their diameter is more than 5 cm and 'giant' when diameter exceeds 15 cm. Hence, Giant POCs are rare and uncommon.[2] They are labelled as paraovarian or paratubal cyst depending on the proximity to the respective structure, though it is observed that the two terms are used interchangeably. When a POC is located at the fimbriated end of the fallopian tube, pedunculated and smaller than 2 cm, it is called as cystic hydatid of Morgagni.[5]

In a large study based on laparoscopic evaluation it concluded that that cysts were paratubal in 40% of patients and paraovarian in 60%. They were unilateral in 67.7% and bilateral in 15.3% with more than one small cyst occurring on one side in the remaining 17% of patients.[6] These cysts originate from the mesothelium of the broad ligament in 68% of cases, rest 30% originate from remnants of paramesonephric duct and from the mesonephric duct remnants in the remaining 2% of

cases.[2]The mesonephric ducts start developing between 20 and 30 days of gestation, contributing to the formation of the male reproductive excretory system, including the vas deferens, epididymis, and seminal vesicles. In females, these ducts typically remain as vestigial structures, often found in the broad ligaments. Occasionally, parts of the epithelial lining may remain unusually active and continue to proliferate, leading to the formation of cystic masses.[7] To be precise they arise from epoophores, which are located in the broad ligament and consist of a longitudinal ductulus and 10-20 transverse ductuluses, and all these ductuli are secretory.[8] Paramesonephric cysts have a hormonal influence which is the reason for their growth and prevalence in reproductive age group and explains their rapid growth in pregnant women.[5,9]

Paraovarian cysts in most cases are asymptomatic and are an incidental finding during pelvic examinations or surgery. More than half of POCs is misdiagnosed as ovarian cysts, tubal cysts, peritoneal inclusion cysts and even mesenteric cysts.[2] However, this case intends to report Bilateral paraovarian cysts with giant on one side and incidental on the other.

CASE REPORT

A 26-year-old P0L0, married for 5 years presented with lump per abdomen since 2 years. Associated symptoms became more bothersome and worrisome as they manifested mainly as feeling of abdominal and pelvic heaviness. There was no anorexia or weakness. Her family and medical histories were unremarkable. Menstrual cycles were irregular with no dysmenorrhea. On presentation her PR was 76 bpm, and BP 122/72 mmHg. General examination was unremarkable. On per abdominal examination, firm and uniformly distended abdominopelvic mass extending from the pubic symphysis to xiphoid process, of 36weeks gravid uterus size, mobile from side to side, non-tender, with smooth margins, regular in contour, dull on percussion with central position of umbilicus and intact hernial sites. No signs of peritoneal irritation and no local rise in temperature was noted.

Laboratory tests revealed HB-13.3g/dl, ovarian tumor markers i.e., AFP, CA19.9, CA125, LDH and Beta HCG were all within normal limit except CEA which was raised at 41.68 ng/ml. The remaining investigations urine routine, Liver function test, Renal function tests and serum electrolytes were within the normal range. Ultrasonography whole abdomen was suggestive of a cystic lesion of 45×50×10 cm with internal reticulations and echoes, suggesting? para ovarian? mesenteric cyst. The cyst had a well-defined regular wall, with a thickness of up to 3 mm, and no evidence of solid components or echogenic contents within. Cystic lesion appeared separate from the ovaries. The ovaries and uterus were also of normal size with an endometrial thickness of 4 mm. CECT pelvis was done considering the financial constraints of the patient which was suggestive of large heterogenous fluid density cystic lesion measuring approximately 50×45×10 cm with wall enhancement and fat composition in abdomen and pelvic cavity, both ovaries were visualized separate from lesion? neoplastic There was no ascites and lymphadenopathy, and other abdominal viscera were normal.

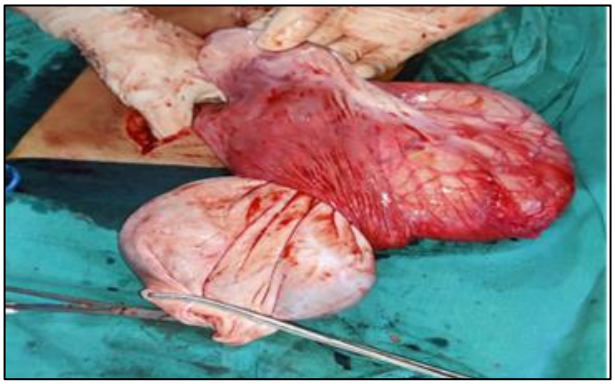

Figure 1: Giant left paraovarian cyst 50×40×10 cm with stretched out fallopian tube and cyst wall.

Figure 2: Giant paraovarian cyst aspirated around 4 litres of straw-coloured fluid which, further allowed POC to be exteriorized with ease.

Figure 3: Right sided paraovarian cyst 5×6 cm found incidentally on exploring the opposite adnexa.

Exploratory laparotomy was planned and left Paramedian incision was taken below the umbilicus. Giant left

paraovarian cyst up to epigastrium with 50×45×10 cm found. Decision was taken to aspirate the fluid contents while avoiding spillage of fluid into the peritoneal cavity. Four litres of straw-coloured serous fluid was aspirated. Following fluid evacuation, the cyst wall was decompressed and collapsed thus it became easier to further exteriorize it from a small incision. Enucleation of the cyst was done with preservation of ovary and fallopian tubes. Careful exploration of the opposite side revealed right sided paraovarian cyst 5×6 cm was aspirated and excised. Right fallopian tube was buried deep within the cyst and adhered to it. Cyst wall was sent for frozen section which revealed benign serous cystadenoma. Fluid aspiration yielded four litres of straw-coloured fluid, which was sent for fluid cytology, was negative for malignancy. Histopathology report of cyst wall was suggestive of paramesonephric or paraovarian benign cyst. The patient's postoperative course was uneventful, and she was discharged 8 days after surgery.

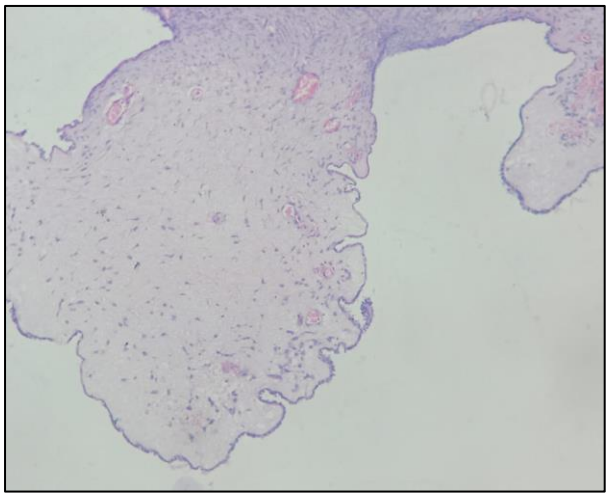

Figure 4: Fibro collagenous wall with foamy histiocytes in sub epithelium.

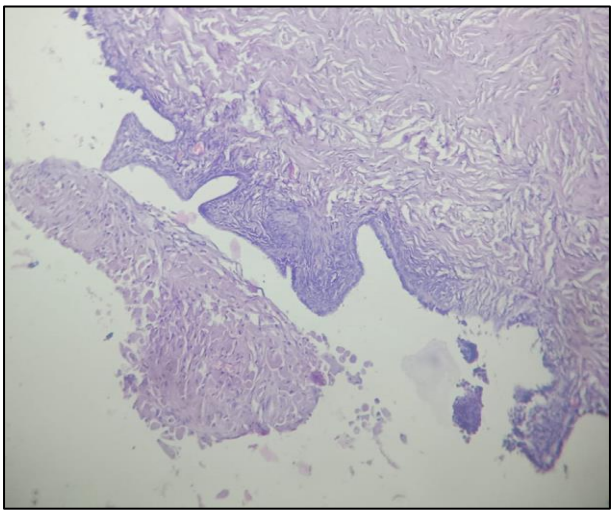

Figure 5: Fibro collagenous wall with foamy histiocytes in sub epithelium.

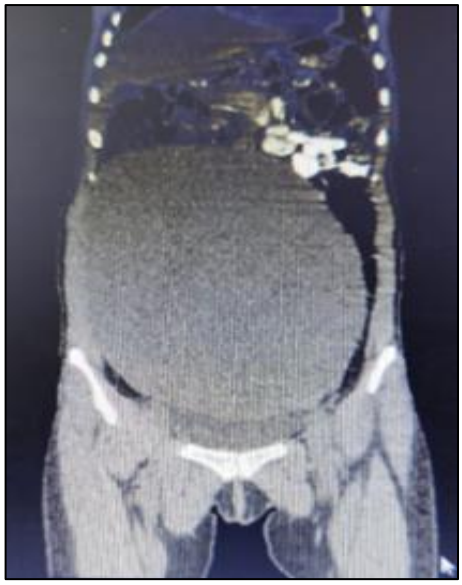

Figure 6: CECT Abdomen pelvis Coronal view.

Figure 7: CECT Abdomen pelvis sagittal view.

DISCUSSION

Paraovarian cysts should be kept in mind as a differential diagnosis for patients with adnexal masses by gynaecologists and radiologists. Despite the high prevalence of POCs and the availability of advanced imaging techniques in modern medicine, an accurate pre-surgical diagnosis is still made in less than 50% of patients.[2] Giant cysts pose a diagnostic dilemma as there is a concern for malignancy, adequate removal demands, and appropriate excision estimation, ensuring preservation of future fertility of certain patients, all the while considering least risk to the patient. Though it is mandatory to test the

values of the serum tumor markers i.e., CA 125, carcinoembryonic antigen (CEA), lactate dehydrogenase (LDH), alpha-fetoprotein (AFP), beta-human chorionic gonadotropin (β HCG). But their values are elevated in only 54% of the adnexal malignant neoplasms and in 6.5% of the benign lesions. Currently, there are insufficient studies on the predictive value of CA 125 levels for malignant POCs.[5]

Ultrasonography is considered as the first-line imaging technique for distinguishing between benign and malignant adnexal masses as it can accurately diagnose POCs in 87.5% of cases. However, this technique is highly subjective based on the expertise of the examiner. As POCs appear as simple cysts by ultrasound and are indistinguishable from ovarian cysts if one does not appreciate its extraovarian location. Occasionally, POCs can have internal echoes due to hemorrhage.[7]

Hence, a correct diagnosis prior to surgery can be made by appreciating ultrasound findings such as the split sign, which is separation of the cyst from the ovary on applying probe pressure and the absence of the rim sign which is appearance of normal ovarian follicles surrounding a cyst.[2] After visiting data from several studies, we came to the proposition that generally POC's appear unilocular, anechoic, thin-walled, with positive splitting sign and an absent rim sign. Also, ovarian cysts have a positive rim sign, an absent splitting sign with the normal ovary not visualized separately from the cyst. Lastly peritoneal inclusion cysts appear irregular, having beak-like margins, with positive flapping sail sign which is appearance of multiple thin oscillating septations.[2] The differential diagnosis of uncomplicated POCs includes simple ovarian cyst, peritoneal inclusion cyst, mesenteric cyst, abdominal lymphangiomas, pancreatic pseudocyst, echinococcal cyst, cystic intestinal duplication, or cystic mesothelioma and hydrosalpinx and malignancy.[5,7]

Clinical features such as rapid cyst growth, weight loss, anorexia, ascites and lymphadenopathy combined with sonographic findings can serve as indicators of malignancy. Ultrasound findings regarding malignancy include presence of papillary or solid components, presence of ascites, an intra tumoral high colour Doppler flow and presence of mural nodules, they are more pertaining to malignancy than other findings like cyst size greater than 10 cm and cyst wall irregularities.[2,4,7] When the mass is large or cannot be visually separate from the ovary an additional MRI is necessary. Ultrasound may not show accurately the origin of this lesions whereas, computed tomography or MRI are more useful in showing a clear delimitation between the normal affected-side ovary and the unilocular cystic lesion.[1]

Complications can present with acute abdominal pain in the case of cyst rupture, intracystic haemorrhage, cyst torsion or perforation. Torsion is more commonly reported on the right side, likely due to the sigmoid colon limiting the cyst mobility on the left side. As POCs do not have a pedicle of their own, their torsion usually involves the ovary, fallopian tube or the infundibulopelvic ligament. Long term complications include an increased incidence of ectopic pregnancy and infertility as they cause tubal narrowing and disturbed tubal motility.[2] Overall the preoperative differentiation between paraovarian cysts and ovarian cysts remains very difficult to be establish and in most of the cases the precise diagnosis is made only during the surgery as according to study by Brahmana et al only 1 in 15 patients are diagnosed preoperatively.[1,10]

Often management of POCs depend upon the presence and severity of the symptoms, the cyst size and ultrasonography characteristics, CA 125 results, age of the patient and the risk of malignancy. Smaller cysts may be aspirated, but there is a high risk of recurrence. POCs of >10 cm diameter or symptomatic cysts should be managed surgically.[5,11,12] Whereas, POCs larger than 30 mm should be excised, due to their constant growth, torsion risk and chances of developing into malignant tumors.[11] Malignant changes have been reported in about 2% to 3% cases.[7] But we must assert ourselves that age, gross appearance, size, septation, and Ca 125 levels are all weak indicators of malignancy. Hence, frozen sections should be checked intraoperatively to confirm the malignancy status especially when there is a papillary projection inside the cyst.[10] The primary goal of treatment should be to preserve ovarian tissue to maintain fertility. While cystectomy is the standard treatment, removing a giant POC may occasionally necessitate associated tubal excision or even oophorectomy.[2,5] Aspirating the cyst content is mandatory for an accurate diagnosis and for an easier cyst dissection.[5]

The final confirmatory diagnosis is by macroscopic and microscopic tissue examination. Histopathological examination may reveal secretory and ciliated cells of paramesonephric origin, low cuboidal epithelium and occasional clear cells of mesonephric origin, and flattened epithelium with occasional tubal differentiation and surrounding fibrous tissue of mesothelial origin.[8] The most frequent reported type was paramesonephric variant in a study by Christina et al.[1]

POCS are lined by a secretory epithelium responsible for cyst formation usually of the serous or mucinous subtypes.[5,12] Hence, great distention of the cavity often leads to distortion of the epithelium and absolute differentiation is difficult of the exact histological variant of POC.[8] Intraoperatively, careful exploration should be performed of both the sides of adnexa to confirm the origin of the cyst, prevent diverting to misdiagnosis, or exploring any finding missed by imaging. This case presented with a rare bilateral paraovarian cyst with one being giant obscuring the presence of the other one.

CONCLUSION

Ovarian lesions are the first differential in case of giant mass per abdomen. Inspite of advanced diagnostic modalities like MRI and CECT, the diagnosis of

paraovarian cysts is generally missed. There are no standard criteria for differentiating between ovarian and paraovarian cysts. We also need to report more of such cases, so that exact incidence of such cysts in known. The diagnosis of giant paraovarian cyst should also be considered while dealing giant mass per abdomen, as it is also associated with the tubal involvement and further reproductive outcome of the patient.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Gadre S, Chaudhary G, Khan FF. A rare case of bilateral paraovarian cysts, giant on one side. Int J Reprod Contracept Obstet Gynecol 2024;13:2532-6.