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A 27-kg Giant Ovarian Mucinous Cystadenoma in a 72-Year-Old Postmenopausal Patient: A Case Report

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

ABCDEFG 1 Lama Nathem Akhras*
ABCDEFG 1 Lana Nathem Akhras*
ACDEFG 1 Saifudheen Faroog
ADEFG 2 Lamiaa AlSebay

1 College of Medicine, Alfaisal University, Riyadh, Saudi Arabia
2 Department of Obstetrics and Gynecology, Specialized Medical Center Hospital, Riyadh, Saudi Arabia

* Lama Nathem Akhras and Lana Nathem Akhras are co-first authors

Corresponding Author: Lama Nathem Akhras, e-mail: lakhras@alfaisal.edu
Conflict of interest: None declared

Patient: Female, 72
Final Diagnosis: Benign ovarian mucinous cystadenoma
Symptoms: Difficulty breathing and ambulation • heartburn • reflux • early satiety • nausea
Medication: —
Clinical Procedure: Total abdominal hysterectomy along with bilateral salpingo-oophorectomy
Specialty: Obstetrics and Gynecology

Objective: Rare disease

Background: Mucinous cystadenoma is a benign cystic ovarian tumor arising from the surface epithelium of the ovary; it usually presents with vague, unspecific abdominal symptoms. If not detected early, they have the potential to grow to a substantial size and can present with huge abdominal distention leading to various compression symptoms. Mucinous cystadenomas most commonly occur in the third to sixth decades of life, and rarely occur in extremes of age. The reported incidence of giant ovarian cystadenoma in postmenopausal women is low or relatively unknown due to widespread use of ultrasound and other radiological imaging modalities these days. Here, we report a case of giant mucinous cystadenoma in a 72-year-old postmenopausal woman with multiple comorbidities.

Case Report: We present the case of a 72-year-old postmenopausal high-risk patient who presented with a huge abdominal distention which started gradually 1 year before. Abdominopelvic ultrasound showed a left giant multiloculated abdominal cyst. An intact 27-kg ovarian cyst was removed, and a total abdominal hysterectomy (TAH) along with bilateral salpingo-oophorectomy (BSO) was performed. The final histopathological report showed a benign ovarian mucinous cystadenoma.

Conclusions: Mucinous cystadenoma is a benign neoplastic disease that can reach a massive size. They are rare in the postmenopausal age group, but when they do occur, they pose a diagnostic and therapeutic challenge. This case report highlights the importance of early detection and management of adnexal masses in postmenopausal high-risk patients to decrease preoperative and postoperative complications and improve quality of life.

MeSH Keywords: Abdominal Neoplasms • Ascites • Case Reports • Ovarian Cysts • Postmenopause

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Background

Ovarian tumors are the most common gynecological neoplasms, with a prevalence of 2.5–6.6% in a woman's lifetime, and are the 8th leading cause of cancer mortality among women worldwide [1,2]. Ovarian neoplasms present in a variety of subtypes and origin of tumor cells, with surface epithelial-stromal cell tumors being most frequent at 65–70% [3]. Among them, mucinous cystadenoma is a benign cystic ovarian tumor originating from the surface epithelium of the ovary, characterized by mucin production and classified into benign (80%), borderline or low malignant potential (10%), and invasive (10%) subtypes [3]. Mucinous cystadenomas occur most commonly in third to sixth decades of life, but rare cases in younger and older women have also been reported [3]. Benign mucinous secreting tumors account for 10–15% of all ovarian neoplasms [4]. Borderline subtypes tend to be more common than invasive tumors, accounting for 67% of mucinous neoplasm versus less than 2.4% in primary mucinous carcinomas [4]. Ovarian cysts are usually asymptomatic at very early stages, until they grow in size [5]. Most ovarian tumors are now detected on ultrasound and other radiological imaging due to their widespread use [6]. Mucinous cystadenomas are unilateral in 95% of cases and can present with vague symptoms like progressive abdominal distention, fullness, bloating, an adnexal mass, vague pelvic or abdominal pain, and gastrointestinal symptoms [5]. If mucinous cystadenomas are untreated, they can grow to gigantic sizes. The aim of this case report and literature review is to highlight such a case in a surgical high-risk postmenopausal woman, which is very rare, and to further expand the literature review in this age group.

Case Report

A 72-year-old multiparous postmenopausal woman presented to our Emergency Department with complaints of massive abdominal distention which started gradually 1 year ago. This was associated with recurrent heartburn, reflux, early satiety, nausea, and, more recently, with difficulty in breathing and ambulation due to rapid increase in girth. There were no other gastrointestinal, urinary, or gynecological symptoms. Initially, she sought medical attention at another center for her symptoms, but did not follow up and assumed she was getting obese.

She was a grand multiparous woman (parity 12, abortions 2) who had a significant past medical history of asthma, hypertension, and type 2 diabetes mellitus. Her past surgical history included 3 cesarean sections, tubal ligation, reversal of ligation, 3 hernioplasties, cholecystectomy, L4 and L5 fixation, and cardiac catheterization. Her family history is positive for malignant ovarian and breast cancer in first-degree relatives.

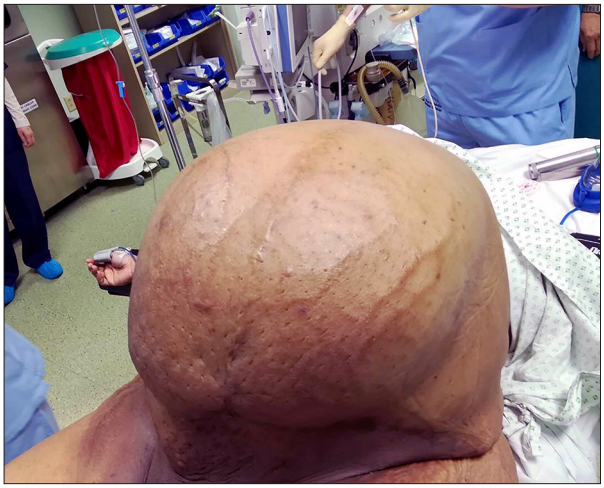

Figure 1. Gross picture of abdomen showing massive distention.

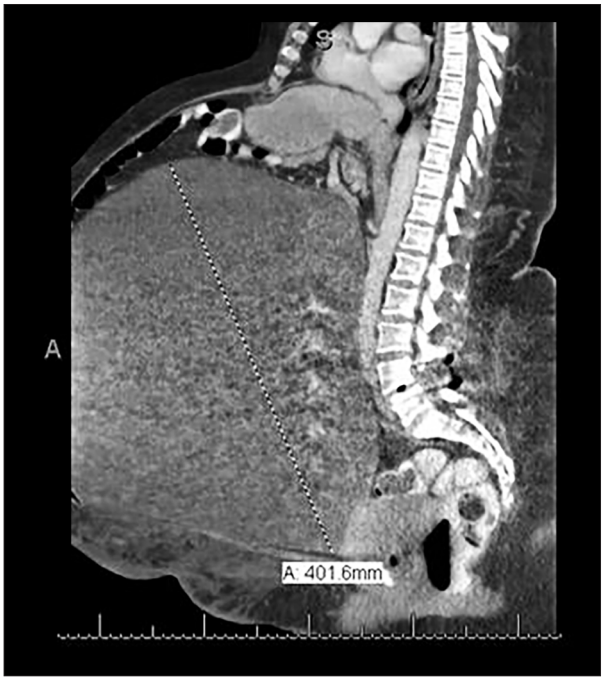

Figure 2. CT scan showing a huge pelvic-abdominal cyst measuring 40×45 cm.

On physical examination, the patient was conscious, alert, and oriented, but in moderate discomfort. Her abdomen was massively distended, with visible skin changes, which included pitting skin discoloration and striae. She was morbidly obese and weighed 160 kg, height 153 cm, and BMI 68.3, with an abdominal girth of 168 cm at the pubic symphysis. The abdomen was grossly distended, hard, non-tender, and tense on palpation (Figure 1). A previous surgical scar on the left lower quadrant was noted. Percussion notes were dull over the abdomen, the fluid thrill and shifting dullness was negative, and bowel sounds were difficult to appreciate due to the large size of the mass. Her vulva, vagina, and cervix were grossly normal.

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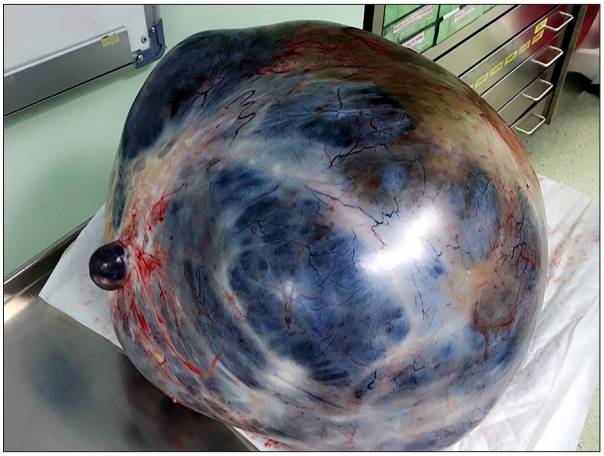

Figure 3. Gross pathology of the tumor. Postoperative cyst weighing 27 kg.

Laboratory investigation showed that hemoglobin was low, at 10.62 g/dl. Eosinophils were elevated (4.05%), and the rest of the CBC was within normal limits. The coagulation profile was within the normal range. All tumor markers were within normal limits. Renal profile and hepatic profile were within normal limits. An ultrasound scan showed a huge multiloculated pelvi-abdominal cyst measuring 25 cm in the anterior-posterior dimension, with septations, internal echoes, and multiple peripheral small cysts, suggesting an ovarian cyst. A CT scan with contrast was done, which found a huge pelvi-abdominal cyst measuring more than 40×45 cm (Figure 2).

Due to her multiple comorbidities, she was assigned an ASA score of III (American Society of Anesthesiologists physical status, ASA PS) and was designated by the cardiologist to be at moderate risk for cardiovascular events. This surgery was considered high-risk due to her age, obesity, multiple previous abdominal surgeries, and restricted daily activities.

Considering all of these findings, after obtaining consent to perform a high-risk surgery, total abdominal hysterectomy with bilateral salpingo-oophorectomy (TAH & BSO) was performed under general anesthesia in a supine position with a midline laparotomy incision. There was massive edema in the anterior abdominal wall layers. A previous mesh was noted with a loop of small bowel adherent to it at the lower 1/3 of the incision, leading to an inevitable small bowel injury. After opening, the left ovary with the cyst were removed intact without an intraperitoneal rupture or spill through multiple gentle manipulations. The resected cyst measured 80×60×30 cm and weighed 27 kg. Then, TAH and right oophorectomy were done. No lymph node involvement was noted. A histopathologist reviewed the case immediately and declared it to be benign; therefore, no staging was performed. A bowel perforation at the site of the incision was repaired using 2-stapler gastrointestinal anastomosis. The patient left the OR in stable condition and recovered uneventfully, with a 30-kg weight loss by the 3rd postoperative day.

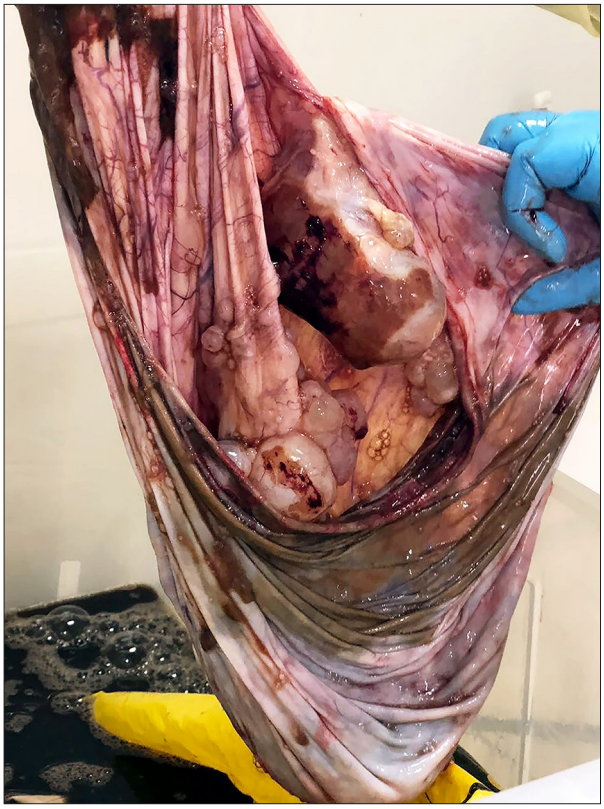

Figure 4. Gross pathology showing cyst shrinkage after dissection. The cyst is filled with viscous hemorrhagic mucinous material.

On gross pathology, when the cyst was resected, it was filled with viscous hemorrhagic mucinous material (Figure 3), and 10 kg of this hemorrhagic mucinous fluid was expelled (Figure 4). Furthermore, large multiloculated cysts with smooth inner walls that are coated with viscous mucoid material were visible. A histopathological examination using hematoxylin and eosin staining of sections reveals a cyst lined by columnar non-ciliated epithelium with abundant mucin with fibrous stroma (Figure 5A–5C). There was no evidence of malignancy. Congested blood vessels were visible, due to the large size of the cyst. Based on all of these findings, a diagnosis of benign mucinous cystadenoma was made.

Discussion

Ovarian neoplasms present in a variety of subtypes and origin of tumor cells. They can arise from surface epithelium, germ cell, or sex cord-stromal tissue. Epithelial cells account for 95% of gynecological malignancies [5]. Overall, mucinous secreting tumors represent 15% of all ovarian tumors [7,8]. In contrast,

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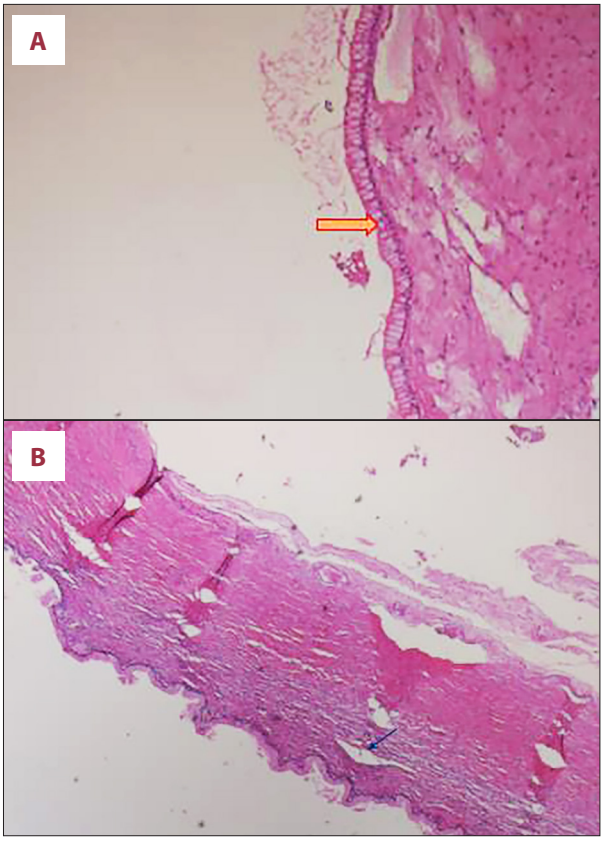

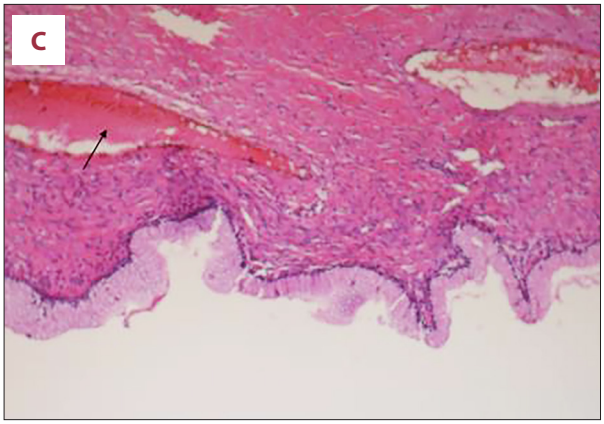

Figure 5. (**A**) (4×) H&E stain section showing benign columnar non-ciliated epithelium of the cyst (orange arrow). (**B**) (10×) H&E stain section showing shows abundant mucin (blue arrow). (**C**) (10×) H&E stain section showing congested blood vessels (black arrow).

serous tumors are the most common ovarian epithelial tumors, in which borderline and malignant serous tumors account for about 60% of all ovarian cancers [9].

Mucinous cystadenoma is a benign cystic ovarian tumor arising from the surface epithelium of the ovary. It is lined by mucin-secreting epithelium. The peak incidence of mucinous cystadenoma is between the third and fifth decades of life (45 years to 65 years) [10]. Histopathological subtyping of mucinous tumors can be classified as benign, borderline or low malignant, and invasive. Overall, 80% are benign, 10% are borderline, and 10% are malignant. Compared to mucinous tumors, serous tumors are more likely to be malignant; 60% are benign, 15% are borderline, and 25% are malignant [9].

Although mucinous cystadenoma is a benign growth, it can grow to gigantic sizes, and the largest reported (in 1963) weighed 148.6 kg [11]. They are usually detected incidentally on imaging studies or during routine gynecological examination [12]; therefore, the reported incidence of giant ovarian cystadenoma in postmenopausal women is low or relatively unknown due to the advanced imaging modalities now available [13]. If symptomatic, they usually present with non-specific symptoms such as abdominal fullness, bloating, pain, or other gastrointestinal symptoms [13]. Common complaints among these patients are related to compression symptoms such as dyspnea, early satiety, heartburn, and increased urinary frequency, which our patient also presented with. Severe complications of ovarian neoplasms include torsion, hemorrhage, and rupture of the adnexal mass, or even death [9]. Specific acute life-threatening complications include pleural effusion and small bowel obstruction, and venous thromboembolism may occur [5]. Upon presentation, the physical findings of our patient were more consistent with an adnexal mass rather than ascites; the abdomen was hard on palpation, with no shifting dullness.

The timeframe to seek medical treatment varies in patients with this tumor due to different presenting symptoms, their willingness to seek medical attention, and their economic status. In some patients the diagnosis can be overlooked because of pronounced obesity or self-neglect in patients who present at a late stage [14]. In addition, fear and anxiety of being diagnosed with cancer can be another factor, as seen in our patient. Ideally, women with high-risk family history of ovarian cancer should be offered early screening with transvaginal ultrasound and serum CA-125, which could have detected this benign adenoma earlier in our patient [15].

Pelvic ultrasound is the imaging study of choice for the evaluation of any adnexal mass [6]. Tumor markers also play a significant role in the initial assessment and follow-up, among which, CA 125 is the most widely used marker, with a sensitivity of 69% to 87% and specificity of 89% to 93% in postmenopausal women [6]. However, this can also be elevated in benign conditions such as endometriosis, adenomyosis, and PID [16]. However, our patient's AFP, CEA, and CA 125 were within normal limits, and the calculated Risk of Malignancy Index (RMI) was 150, which is below the cutoff value of 200 [17]. RMI has been proven statistically to be effective in discriminating

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between benign and malignant masses, and it can be calculated using the product of the serum CA 125 level (U/ml), the ultrasound scan result (expressed as a score of 0, 1, or 3), and the menopausal status (1 if premenopausal and 3 if postmenopausal) [17]. Using an RMI cutoff level of 200, the sensitivity was 85% and the specificity was 97% [17].

There is currently insufficient evidence to support the routine clinical use of other tumor markers such as human epididymis protein 4 (HE4), carcinoembryonic antigen (CEA), CDX2, cancer antigen 72-4 (CA72-4), cancer antigen 19-9 (CA19-9), alpha-fetoprotein (AFP), lactate dehydrogenase (LDH), or beta-human chorionic gonadotropin (HCG) to assess the risk of malignancy in postmenopausal ovarian cysts [18]. According to a report by Brown and Frumovitz, BRCA and p53 mutations are infrequent in mucinous tumors of low malignant potential; thus, these markers were not tested in our patient [4]. Size and laterality are important contributors to the diagnostic course of the tumor origin. Primary mucinous tumors are more likely to be unilateral and larger (5–48 cm) compared with metastatic lesions (2–24 cm) [4]. CT scan is preferred in preoperative assessment of patients with suspected or known ovarian cancer to better plan the surgery [19].

Postmenopausal women are at a higher risk of gynecologic malignancy; thus, any adnexal mass in this age group should be approached cautiously. Therefore, the American College of Obstetrics & Gynecology (ACOG) recommends adnexal masses with suspicious malignant features like ascites, elevated CA125, and evidence of metastasis to be referred to a gynecological oncologist. Studies have shown a 6- to 9-month median survival benefit when a gynecological oncologist performs staging and cytoreduction compared with an operation performed by a general gynecologist [19]. Although our patient did not have any of these features, she was still considered a high-risk patient due to her comorbid conditions and time of presentation.

Ultimately, surgery with intraoperative pathological evaluation remains the criterion standard of treatment for a large ovarian mass [20,21]. The minimally invasive laparoscopic approach is increasingly used as it has been associated with a shorter recovery and decreased perioperative morbidity compared with laparotomy. Moreover, this has been successful even in cases of large ovarian mass [22,23]. However, laparotomy is preferred by most surgeons for very large masses and those suspicious of malignancy. Surgeons should be careful to remove the ovary intact without spillage to minimize the risk of recurrence [24]. In our case, we were able to remove the tumor intact without the need for intraoperative drainage. To reduce the risk of other malignancies, a total hysterectomy and bilateral salpingo-oophorectomy are usually considered in postmenopausal women, regardless of the histology [12]. Concurrently, we performed it on our patient after obtaining consent to perform high-risk surgery.

Hemodynamic instability has always been a feared complication while operating on a huge adnexal mass due to its compression of the vena cava, thus reducing venous return [25]. In addition, splanchnic vasodilation after the mass removal causes further instability [25,26]. Pulmonary edema has also been reported, probably due to the sudden expansion of the chronically collapsed lung [27,28]. To reduce such complications, intraoperative drainage has been recommended [29]. Many case reports have shown that slow removal can adequately prevent such hemodynamic instability, which we also demonstrated in our case by gently removing the tumor.

Conclusions

Mucinous cystadenoma is a benign neoplastic tumor that can reach a massive size. They are rare in the postmenopausal age group, and when they do occur, they pose a diagnostic challenge to differentiate from a malignant neoplasm. Moreover, the patient's multiple co-morbidities, older age, and size of tumors can make the surgical treatment high risk and could lead to a broad set of complications. This case report highlights the importance of early detection, preoperative evaluation, and subsequent surgical management with a multidisciplinary approach in order to decrease preoperative and postoperative complications and improve the patients' quality of life.

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Conflicts of interest

None.

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