Case Report

ANNALS OF MEDICINE & SURGERY

OPEN

A case report of bilateral benign Ovarian Fibrothecoma coincidental with ascites: an unconventional co-occurrence

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Introduction and importance: Ovarian fibrothecoma are a mostly benign and profoundly rare type of gonadal stromal cell tumor. It makes up 3–4% of all kinds of ovarian neoplasia. They are predominantly unilateral in origin and mostly present in women during the postmenopausal phase. Our case is especially important because the tumors occurred bilaterally and were associated with ascites. This seldom happens in patients with ovarian fibrothecoma. Early identification and treatment are key components to avoid the subsequent complications of this tumor.

Case presentation: We present the case of a 54-year-old female who presented complaining only of a slow progressive increase in the abdominal contour associated with vague abdominal pain. Our preoperative radiological imaging revealed multiple ovarian and uterine masses.

Clinical discussion: Surgical intervention in the form of a hysterectomy with bilateral salpingo-oophorectomy was achieved. Histopathological analysis revealed bilateral benign ovarian fibrothecoma with benign uterine leiomyomas. The patient underwent an uneventful postoperative recovery.

Conclusion: Ovarian Fibrothecoma is a rare gynecological pathology. The uniqueness of our case stems from the rarity of its bilateral occurrence and in rare occasions, their occurrence is accompanied by ascites. This kind of co-occurrence should be differentiated from other rare presentations, such as Meigs Syndrome. Therefore, documentation is necessary to circumvent misdiagnoses and to abate the resulting patient morbidity. To further highlight the value of our case, it is to the best of our knowledge, the first documented case of this pathology from our country.

Keywords: ascites, bilateral benign ovarian fibrothecoma, case report, gynecological neoplasia, ovarian tumors, pelvic surgery

Introduction

Ovarian neoplasia has many forms, some benign and others malignant. Sex cord-stromal neoplasia is considered a rare and distinguished type of ovarian tumor. They account for almost 8% of all kinds of ovarian neoplastic entities. This class of tumors is considered gonadal cell types and is either derived from mesenchymal cells or the coelomic epithelial cell layer during embryonic gonadal development. One of the three most prevalent types of these tumors is fibrothecoma [1]. Ovarian fibrothecoma is a considerably rare class that is of gonadal stromal cell origin, and they comprise 3–4% of all kinds of ovarian neoplasia[2,3]. They are mostly benign and therefore are rarely malignant[4,5]. They occur unilaterally in 90% of the cases and mostly in women during the postmenopausal period[3,6]. In comparison with the previous information, our case involved a bilateral, rather than unilateral, ovarian fibrothecoma. Upon careful review of the published literature, we concluded that no previous similar cases from our country were documented. This constitutes a vital medical precedence that warrants documentation. This will result in the timely detection and treatment of affected patients.

HIGHLIGHTS

- Ovarian fibrothecoma are a rare type of gonadal stromal cell tumor. It makes up 3–4% of all ovarian neoplasia.
- They are predominantly unilateral and mostly affect females during the postmenopausal phase.
- Clinical presentation and radiological features are vague. Hence, it can be misdiagnosed with other entities.
- They are rarely accompanied by ascites and mainly occur with a normal CA-125.
- To the best of our knowledge, ours is the first documented case from our country of a bilateral ovarian fibrothecoma.

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Sponsorships or competing interests that may be relevant to content are disclosed at the end of this article

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Annals of Medicine & Surgery (2023) 85:3739–3743

Received 11 May 2023; Accepted 10 June 2023

Published online 21 June 2023

http://dx.doi.org/10.1097/MS9.0000000000001016

The work has been reported in line with the Surgical CAse REport (SCARE) criteria and the revised 2020 SCARE guidelines[7].

Presentation of the case

Patient Information

We present the case of a 54-year-old female with a known history of Diabetes Mellitus Type II and arterial hypertension. She presented to our surgical clinic complaining of a gradual and painful increase in the abdominal contour, in addition to gradual dyspnea for 1 month. The abdominal contour was symmetrically increased and it was simultaneously accompanied by diffuse and vague abdominal pain. The pain had no radiation, its intensity was 03/10 on the patient's numerical pain scale, had no associated symptoms, no specific triggers, and was partially relieved by paracetamol. Symptoms were simultaneously accompanied by progressive dyspnea that was especially noticed upon changing body position, and this was markedly associated with orthopnea. It is worth noting that the patient denied any change in her appetite and her bowel habits. Furthermore, no jaundice, nausea, emesis, or pallor was noticed. Moreover, the patient denied any accompanying cough, chest pain, or palpitations. The patient denied experiencing any B-Symptoms, such as fever, unintentional weight changes, or cold/night sweats. The patient's gynecological history included regular menstruation cycles until experiencing menopause at 44 years of age with no postmenopausal symptoms. There was no previous history of abortions, hormone replacement therapy, or the use of contraceptive medications. Additionally, no genitourinary symptoms were reported, and no history of a previous similar incidence exists. Her drug history involved oral antihypertensive medications and oral antidiabetic agents. Moreover, her family history included benign uterine leiomyomas, whereas her surgical history was unremarkable, and her allergic and psychosocial histories were negative. Her BMI measured 29 Kg/m^2.

Clinical findings

We began our physical examination by taking the patient's vital signs. She was mildly tachycardic and tachypneic. The results of the rest of the examination were normal. Upon inspection, the abdomen was moving symmetrically with respiration, and it was symmetrically enlarged. No jaundice, pallor, spider naevi, caput medusa, or hypo-/hyperpigmentation were seen. Upon palpation, the abdomen was soft and displayed no tenderness or guarding. A considerably large left-sided pelvic mass was palpated. It was hard and mobile with relatively well-defined borders. In addition, signs such as shifting dullness and transmission thrills were positive. By percussion, the abdomen was dull, whereas the auscultation findings included tympany at first then dullness over the area with fluid accumulation. A gynecological pelvic examination elicited a seemingly normal uterine size, and no abnormal discharges or bleeding were present.

Diagnostic assessment

Abdomino-pelvic ultrasound revealed a large amount of ascitic fluid with bilateral poorly demarcated pelvic masses with mixed echogenic properties. These two findings in addition to the patient's postmenopausal status and her CA-125 value earned 1773 points on the Risk of Malignancy Index for ovarian cancer. This means it has a high risk for being malignant. However, further assessment could not be achieved due to a large amount of free fluid. A high-resolution contrast-enhanced Multi-Slice Computed Tomography scan of the chest, abdomen, and pelvis was done. It revealed the existence of multiple soft tissue pelvic masses situated in the uterine and adnexal region. The largest of which was in front of the rectum and measured (6.5×9 cm). The second mass was located superior and anterior to the bladder and measured (8×12.5 cm). The radiological features of these lesions were suspicious of malignancy. Finally, a moderate amount of ascites was present in the perihepatic, perisplenic, in between the bowel loops, and in the pelvis. No frank lymphadenopathy was demonstrated. No pleural/pericardial effusions and no thoracic lesions were seen (Fig. 1 A-B-C-D). A comprehensive laboratory panel was done. The sole anomaly was an increased CA-125 value of 197 U/ml.

Surgical intervention was necessary given the preoperative assessment results. The patient was assigned to a nil-per-mouth nutritional status, had two large-bore intravenous cannulas set up, and was supplied with suitable preoperative antibiotics. We did not encounter any hindrances or challenges during any of the perioperative phases.

Therapeutic intervention

We carried out our surgical intervention at a specialized tertiary hospital. Surgery was achieved by a Gynecology specialist, General Surgery specialist, and a first surgical assistant specialized in General Surgery with 10, 11, 5 years of experience, respectively. General anesthesia was achieved without any reported perioperative complications. Through a vertical midline incision, optimal surgical exposure was achieved. A considerable volume (3L) of ascitic fluid was immediately noticed and was suctioned accordingly. Samples of which were taken for cytological analysis that revealed the presence of reactive mesothelial cells without any abnormal cells. Furthermore, a large left ovarian mass (Fig. 2) was in addition to multiple uterine mass-like lesions and a smaller right ovarian mass were visualized. Based on these findings, a hysterectomy with bilateral salpingo-oophorectomy was done (Fig. 3). Moreover, the greater omentum, a section of the parietal peritoneum, and several regional lymph nodes were resected. The entirety of the excised specimens was directly sent to the pathology laboratory to undergo a thorough histopathological analysis. Results of the subsequent histopathological analysis indicated a bilateral benign ovarian fibrothecoma (Fig. 4 A-B-C-D). Furthermore, the uterine lesions were diagnosed as benign sub-serosal leiomyomas and the analyzed omentum, lymph nodes, parietal peritoneum, and ascitic fluid revealed no malignancy or tumor involvement. The patient's symptoms completely subsided during the postoperative period. She was discharged home on the fifth postsurgical day. The patient has been followed up in our surgical clinic for 8 months so far where she has received periodic physical and radiological assessments to ensure her successful recovery. CA-125 returned to normal levels. She underwent a complete recovery, and the results of the clinical assessments were all within normal. Additionally, she was referred to a specialized oncologist for any additionally needed follow-up protocols.

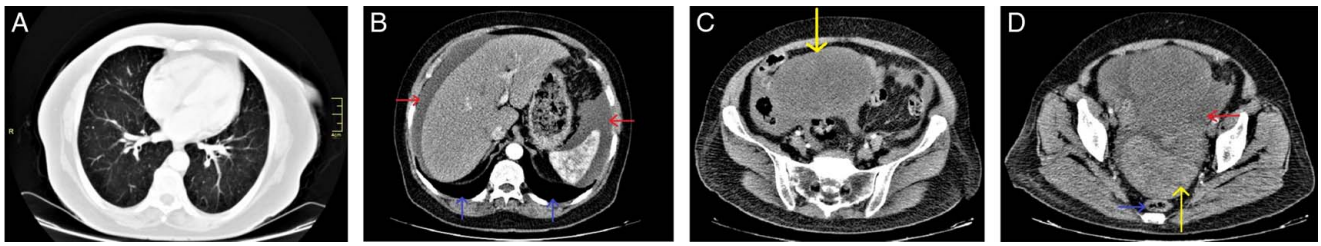

Figure 1. (A) Preoperative cross-sectional view of the Multi-Slice Computed Tomography (MSCT) scan showing a clear lung field with no signs of free fluid accumulation. (B) Preoperative cross-sectional view of the Multi-Slice Computed Tomography scan of the abdomen and pelvis. The *Red Arrows* identify the presence of ascitic fluid, whereas the *Blue Arrows* identify the pulmonary base as it is free of any fluid accumulation. (C) Preoperative cross-sectional view of the Multi-Slice Computed Tomography scan of the abdomen and pelvis. The *Yellow Arrow* indicates a heterogeneous ovarian mass that measured (8 × 12.5 cm). (D) Preoperative cross-sectional view of the Multi-Slice Computed Tomography scan of the abdomen and pelvis. The *Red Arrow* identifies the ascitic fluid, whereas the *Yellow Arrow* identifies a large ovarian mass, and the *Blue Arrow* identifies the rectal segment that seems to be relevantly compressed by the mass.

Discussion

The composition of ovarian fibrothecoma is an intercalating mixture of thecomatous and fibrous components[8]. These tumors predominantly favor occurring in the population of post-menopausal women[3]. In rare cases, they co-occur with Meigs syndrome which involves the presence of ascites, pleural effusion, and high levels of serum CA-125[8,9]. Patients who are affected by ovarian fibrothecoma have a misleading and nonpathognomonic clinical presentation. Presenting symptoms include pelvic pain, the presence of pelvic mass formation, and metrorrhagia. In rare instances, around 8% of patients present with ovarian torsion[9,10]. Preoperatively, establishing the diagnosis of an ovarian fibrothecoma could pose a serious challenge for physicians. This is owed to two reasons; First, due to their extreme rarity, and second, they are commonly misdiagnosed for another uterine lesion like leiomyomas because of their solid nature. Furthermore, the co-presence of ascites, hydrothorax, and high levels of CA-125 result in misdiagnosing ovarian fibrothecoma as a malignant ovarian tumor of another kind[3,11,12]. Sonography is the cornerstone in diagnosing ovarian tumors as well as ovarian fibrothecomas. Variant sonographic features are demonstrated and are particularly comprised of masses that are characterized by an echogenic, mixed echogenic, or hypoechoic appearance[3]. Ovarian fibrothecomas appear on computed tomography as a homogenous solid tumor with diverse levels of radiological enhancement[13]. Generally, serum levels of CA-125 do not raise any red flags because they are typically within normal limits, but in rare cases, high levels could be ensued and resort to normalization after comprehensive tumor resection[14]. When large tumors are present, ascites could simultaneously occur, and it is then suggestive of malignant transformation. Ascites is explained by fluid secretion triggered by the tumor. Tumor secretions bypass the surface layer of the tumor and overwhelm the capability of the peritoneum to resorb these fluids. Another mechanic is the physical exasperation of the overlying peritoneal surface by the mechanical compression effect of the neoplasm[11,15,16]. Multiple differential diagnoses for ovarian fibrothecoma come into consideration. Examples are uterine leiomyomas, and variable types of ovarian lesions like dysgerminomas, and granulosa

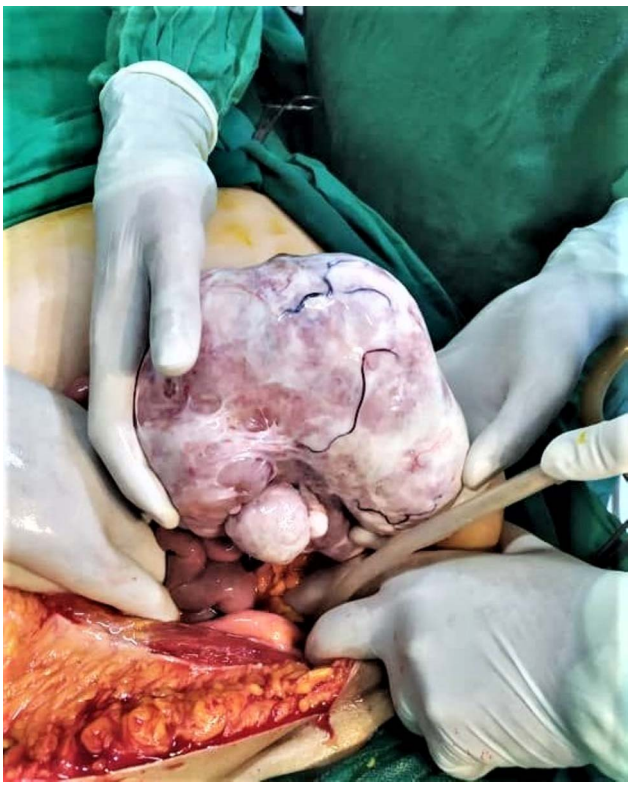

Figure 2. Intraoperative image prior to resection. It depicts the large left ovarian mass.

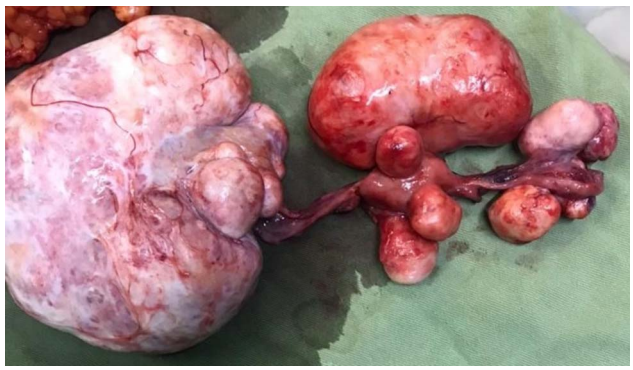

Figure 3. Intraoperative image showing the resected specimens that clearly show the bilateral ovarian masses in addition to the uterine mass formations.

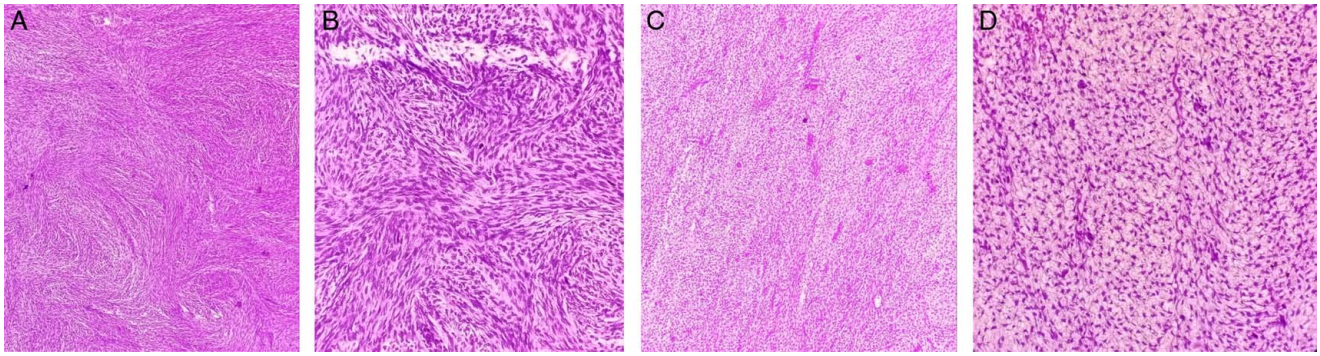

Figure 4. (A-B): Microscopic images (×100 and ×200, respectively) of the histopathological analysis of the bilateral ovarian masses via H&E staining. They demonstrate a morphologic appearance between fibroma and thecoma. In addition, interconnecting collagen fibers and spindle cells aligned in a whirlpool appearance. (C-D): Microscopic images (×100 and ×200, respectively) of the histopathological analysis of the bilateral ovarian masses via H&E staining. They demonstrate the foamy appearance of the cytoplasm which differentiates it from the typical appearance of a Fibroma.

cell tumors[2,4,9]. The earlier the diagnosis is established, the earlier the therapeutic intervention is carried out to save patients from unwanted complications of ovarian fibrothecoma[13]. The gold standard for treatment for younger females is complete resection of the mass, whereas, for older postmenopausal women, it is in the form of bilateral salpingo-oophorectomy[3]. Fibrothecomas are characterized by a white verticillated look on gross examination mimicking the appearance of leiomyomas of the uterus[2]. The neoplasm could appear lobulated or globular and is enclosed by an overall unimpaired ovarian mucosa[9]. On microscopic histopathological analysis, the existence of spindle, oval, or round cells creating numerous quantities of collagen is an identifying trait for this type of tumor[4]. Intracellular lipid exists in a lesser number of theca cells[2].

Conclusion

Ovarian fibrothecoma are a relevantly rare gynecological neoplastic entity. Our case is unique because they rarely occur bilaterally and in seldom cases, their occurrence is associated with ascites. Furthermore, most cases are accompanied by normal CA-125 levels, whereas our case witnessed a markedly elevated level of this marker. This type of pathological co-occurrence should be carefully differentiated from the rare entity known as Meigs Syndrome. Therefore, proper documentation is warranted to avoid misdiagnosis and to minimize the possible patient morbidity. To mark the additional uniqueness of our case, it is to the best of our knowledge, the first documented case of this pathological occurrence from our country.

Ethical approval and consent to participate

Institutional review board approval is not required for deidentified single case reports or histories based on institutional policies.

Consent of patient

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Sources of funding

N/A.

Author contribution

R.M., O.A., R.A.: Conceptualization, resources, methodology, data curation, investigation, who wrote, original drafted, edited, visualized, validated, and literature reviewed the manuscript.
R.M.: First surgical assistant during the operation.
M.S.: Validation of the manuscript.
A.M.: General Surgery specialist who performed and supervised the operation, in addition to supervision, project administration, resources, and review of the manuscript.
M.A.: Gynecology specialist who performed and supervised the operation, in addition to supervision, project administration, resources, and review of the manuscript.
O.A.: The corresponding author who submitted the paper for publication.
All authors read and approved the final manuscript.

Conflicts of interest disclosure

The authors declare that they have no financial conflict of interest with regard to the content of this report.

Research registration unique identifying number (UIN)

N/A.

Guarantor

Omar Al Laham.

Availability of data and materials

The datasets generated during and/or analyzed during the current study are not publicly available because the data were obtained from the hospital computer-based in-house system. Data are available from the corresponding author upon reasonable request.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Acknowledgements

None.

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