Case Report

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# Struma ovarii-associated pseudo-Meigs' syndrome with concomitant abdominopelvic tuberculosis masquerading as ovarian malignancy

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**Abstract:**

Pseudo-Meigs' syndrome (PMS) is a rare disease characterized by the triad of (1) an ovarian neoplasm, other than a fibroma or thecoma, (2) ascites, and (3) pleural effusion. Tumors such as struma ovarii, mucinous and serous cystadenomas, and germ cell tumors have been linked with the condition. Due to its clinical features combined with the elevation of serum cancer antigen 125 (CA-125) levels, it is often mistaken and treated as a malignant ovarian tumor. Ascites or pleural effusion could be massive leading to various life-threatening complications. Despite its presentation, this entity has an excellent prognosis when surgical excision of the tumor is performed. This article presents an unusual case of a 41-year-old gravida 10 para 10 (10-0-0-9) who was diagnosed with a case of struma ovarii associated PMS with concomitant abdominopelvic tuberculosis and elevated CA-125 resembling an ovarian malignancy.

**Keywords:**

Ascites, benign, elevated cancer antigen 125, pseudo-Meigs' syndrome, struma ovarii

## Introduction

Struma ovarii, known as ovarian goiter, is a specialized mature teratoma derived from germ cell layers wherein thyroid tissue (>50%) has predominantly overgrown other elements.[1] This was first described by Bottlin in 1988, and further reports were published by Gottschalk. By 1933, Plaut discovered that thyroid tissues in the mass and thyroid gland are morphologically, biochemically, and pharmacologically identical.

The majority of cases are asymptomatic and are incidental findings during imaging examinations. However, when a multilocular abdominopelvic mass presents with ascites and pleural effusion, it is recognized as pseudo-Meigs' syndrome (PMS). The exact prevalence of PMS is currently unknown, but 5% of this syndrome has been associated with struma ovarii.[1] This extremely rare condition of PMS coexisting with elevated serum cancer antigen 125 (CA-125) can be interpreted as a highly suspicious case of malignancy; hence, causing difficulties in making accurate preoperative and intraoperative diagnoses.[1,2] In such cases, patients are subjected to extensive surgeries for benign conditions because of its malignant presentation.[3] In addition,

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there is no established universally accepted algorithm for the diagnosis and treatment of struma ovarii-related PMS.[4]

## Case Report

A 41-year-old gravida 10 para 10 (10-0-0-9) was consulted at the Obstetrics and Gynecology emergency room with 1-week history of dyspnea secondary to progressive abdominal distention with massive ascites. She has no known comorbidities, no previous surgeries, and no known exposure to any infectious diseases. The family, personal, and social history were noncontributory.

She had her menarche at 12 years old with subsequent menses coming at regular monthly interval lasting for 3–5 days, soaking 2–3 pads per day with no associated dysmenorrhea. Her coitarche was at 18 years old with one nonpromiscuous sexual partner. She took oral contraceptive pills for 4 years. All her pregnancies were carried to term and delivered vaginally at home with no fetomaternal complications.

Eight months before admission, the patient noted a 10 cm × 10 cm hypogastric mass accompanied by gradual abdominal enlargement, anorexia, weight loss, and a reducible perineal bulge. She also developed dull abdominal pain, which prompted consult at a local hospital where a pelvic ultrasound was done revealing an abdominopelvic mass probably ovarian in origin and an elevated value of CA-125 at 600 U/mL (normal value = <35 U/mL); hence, referral to a tertiary institution. Three months before admission, the patient sought consult with a tertiary institution. A transvaginal ultrasound was done, which showed a multiloculated, multiseptated cystic mass measuring 10.3 cm × 11.6 cm × 7.3 cm, malignant by the International Ovarian Tumor Analysis, occupying the abdominopelvic cavity more to the right. There were occasional solid areas, largest measuring 3.7 cm × 3.0 cm × 2.3 cm. Color flow mapping showed moderate peripheral flow with intratumoral flow to solid areas. She was scheduled for preoperative clearance before elective surgical management but due to her progressive dyspnea, she was admitted to the emergency room.

On physical examination, the patient was normotensive at 130/90 mmHg, tachycardic at 140 beats/min, and tachypneic at 26 breaths/min. Oxygen saturation was 96%–99%. She appeared cachectic with bitemporal wasting. Chest findings showed supraclavicular retractions and decreased bibasal breath sounds. Abdomen was globular, distended with an abdominal girth of 129 cm, nontender, with normoactive bowel sounds, and positive for fluid wave test. There was a movable mass at the hypogastric area measuring 10 cm × 6 cm. Pelvic examination showed a prolapsed, irreducible mass with areas of excoriations. The rectovaginal examination was unremarkable. Grade 2 nonpitting bipedal edema was noted on both extremities.

Oxygen was administered at 10 pm through face mask. A cardiac wedge was placed to maintain 45°C high-back rest. Ultrasound-guided paracentesis was done for symptomatic relief. Eighteen liters of serous clear, bright, and yellow ascitic fluid was slowly drained, thereby decreasing the abdominal girth to 120 cm. Peritoneal fluid studies revealed a positive (+1) acid-fast bacilli (AFB) smear and negative cytology. Sputum and stool AFB was negative. Her blood work-up showed hypoalbuminemia. Tumor markers revealed elevated diluted CA-125 at 2,796 U/ml (normal value <35 U/mL) and HE4 at 99.8 pmol/L. The calculated risk of ovarian malignancy algorithm score was 36.75%, hence an increased likelihood for malignancy. Other tests such as liver and kidney function tests, serum electrolytes, and urine analysis were within the normal range [Table 1].

Repeat transvaginal and transabdominal ultrasound reported an increase in the size of the mass to 13.9 cm × 12.9 cm × 10.0 cm with the same characteristics [Figure 1]. Pelvic organ prolapse of the anterior and central compartments was also seen. Furthermore, whole abdominal computerized tomography (CT) scan with contrast indicated 7.7 cm × 10.8 cm × 10.6 cm complex lobulated mass, probably a malignant teratoma in the presence of ascites and left pleural effusion [Figure 2]. The chest X-ray confirmed mild pleural effusion on the left and showed subsegmental atelectasis of parenchymal fibrosis on the right [Figure 3].

**Table 1: Laboratory results**

**A. Peritoneal Fluid Studies**

| Parameter | Result |
|---|---|
| Qualitative | |
| Color | Bright yellow |
| Transparency | Clear |
| Quantitative | |
| Glucose | 7.6 mmol/L |
| Total Protein | 75.06 g/L |
| AFB Smear | +1 AFB Seen (1-29 AFB/20 fields) |
| Cytology | Rare Degenerated Inflammatory Cells |
| Gram Stain | PMN: No Organism Seen |
| Culture & Sensitivity | No growth after 4 days of incubation |

**B. Serum Chemistry and Tumor Markers**

| Test | Reference Values | Results | |
|---|---|---|---|
| | | 1st admission | 2nd admission |
| ALB | 35-50 g/L | 31 | 32 |
| CA125 (diluted) | < 35 U/mL | 2796 | |
| HE 4 | Pmol/L | 99.8 | |

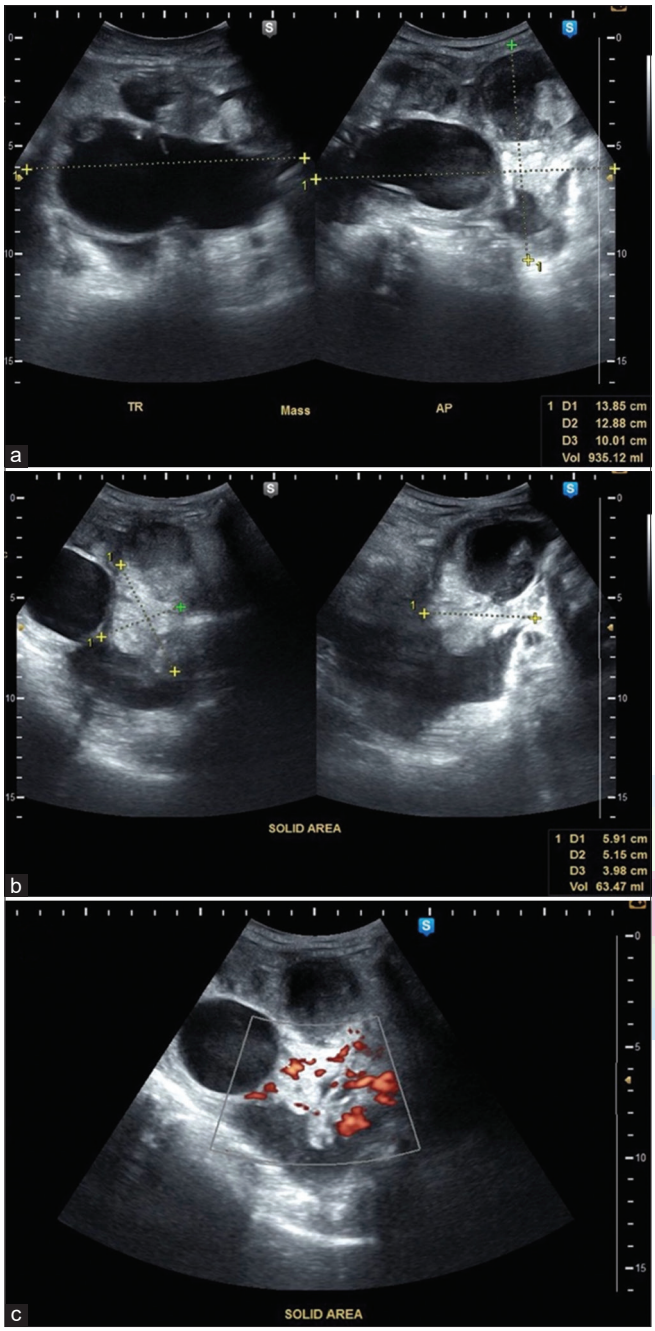


**Figure 1:** Transvaginal and transabdominal ultrasound results showing (a) abdominopelvic mass, (b) solid area of mass, and (c) color flow mapping showing moderate peripheral flow with intratumoral flow to solid areas

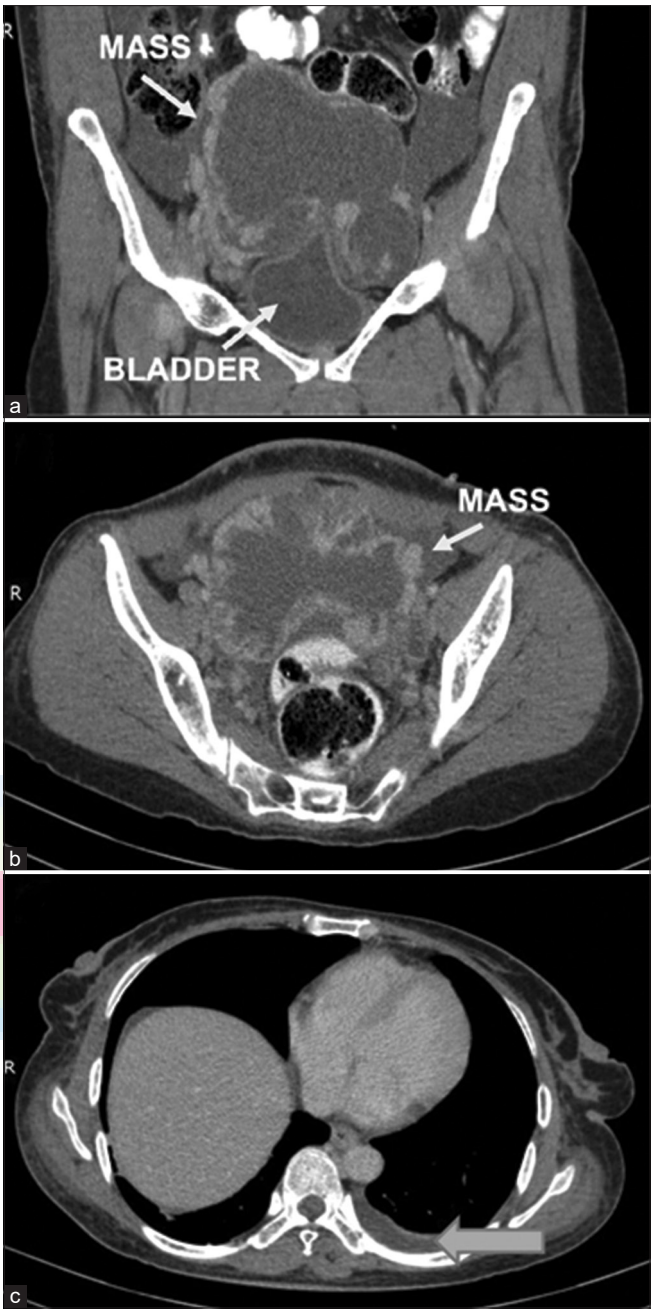


**Figure 2:** Computed tomography scan of (a) abdomen showing a mass above the urinary bladder and central in location, (b) abdomen showing the mass exhibiting variable thickening with enhancement at the walls and nonenhancing cystic appearing at the internal region, and (c) chest showing minimal left pleural effusion

The working impression was ovarian new growth, probably malignant, extrapulmonary tuberculosis (abdominopelvic), and pelvic organ prolapse Stage IV. Referrals to the divisions of gynecologic-oncology, obstetrics and gynecology (OB) infectious disease, and urogynecology were facilitated. The consensus was to initiate anti-Koch's medication before surgery while administering albumin correction. The infectious disease division started Category I tuberculosis treatment (6-month rifampicin containing standardized regimen) and advised completing a month of therapy before surgical intervention. In the interim, her condition improved, and she was sent home with good compliance with her medications. She was subsequently readmitted for definitive surgical management with a note of reaccumulation of ascites and persistence of bipedal edema.

She underwent exploratory laparotomy, total hysterectomy with bilateral salpingo-oophorectomy

with staging procedure, appendectomy, halban culdoplasty, and uterosacral fixation under continuous lumbar epidural anesthesia. After draining 11 l of serous ascitic fluid, a right ovarian tumor was identified. It was a firm, tan-gray to flesh brown, and multilobulated mass, which measured 13.0 cm × 16.0 cm × 6.0 cm with smooth and intact capsule. Cut sections showed multiple complex solid-cystic areas, the largest of which measured 6.5 cm × 3.5 cm × 1.6 cm. The cavities were filled with yellow mucoid material, with no areas of hemorrhage or necrosis. No hair, bones, or sebaceous glands were seen [Figure 4]. The other structures were grossly normal. The final histopathologic results showed a right struma ovarii, (9.5 cm in greatest tumor dimension), with no malignant component [Figure 5]. Other specimens were histologically unremarkable and negative for tumor.

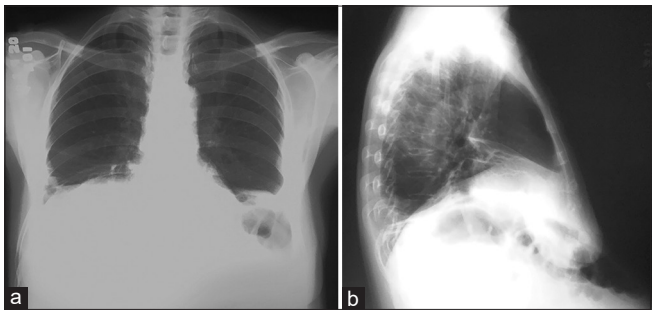

**Figure 3:** Chest radiography showing parenchymal fibrosis, right and pleural effusion, left (a) anteroposterior view and (b) lateral view

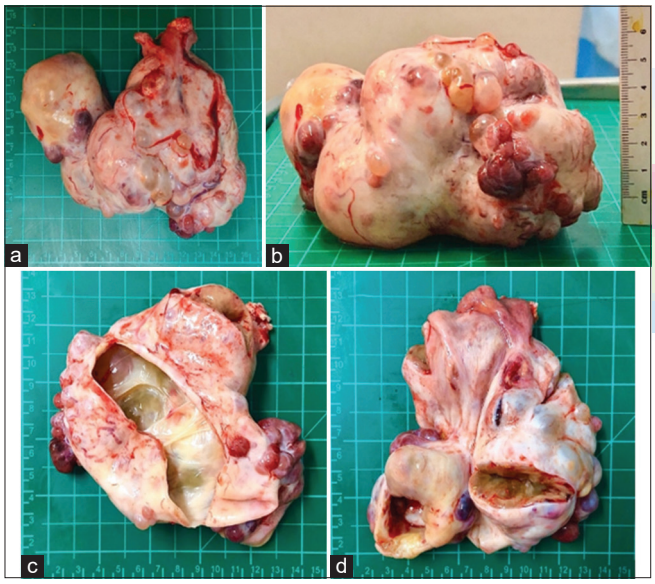

**Figure 4:** Macroscopic findings showing a multilobulated mass measuring 13.0 cm × 16.0 cm × 6.0 cm (a) anterior view, (b) lateral view, (c) cut-section anterior view, and (d) cut-section posterior view

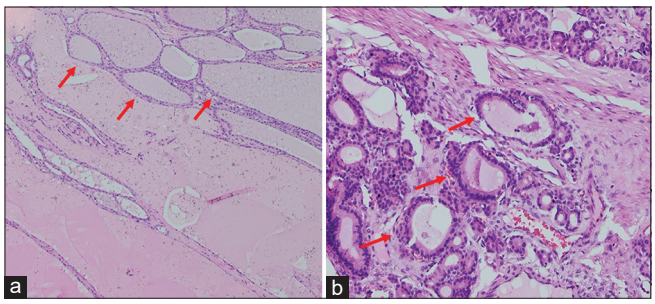

**Figure 5:** Microscopic findings showing tumor with multiple thyroid macrofollicles filled with colloid using hematoxylin and eosin stain (a) low magnification objective, (b) high-power objective

The postoperative course was uneventful. There was a resolution of the ascites and pleural effusion and completion of anti-Koch's medication after a follow-up of 6 months.

## Case Discussion

Struma ovarii is a rare monodermal ovarian teratoma with the presence of thyroid tissue components comprising more than 50% of the mass.[5] It represents 1% of ovarian tumors and constitutes 0.3%–2.5% of ovarian teratomas. This tumor is generally benign, but the malignant transformation has been reported in 5%–7% of cases, and metastatic disease was even more uncommon. Papillary carcinoma is the most frequent type of thyroid malignancy that occurs in struma ovarii.[1] A review of local literature showed two published online journals on struma ovarii. These were cases of papillary thyroid-type carcinoma arising from struma ovarii and struma ovarii with serous cystadenoma, but none involved PMS.

The peak incidence occurs between the fifth and sixth decades of life and is more common in postmenopausal women. Although patients ages 23–72 years old have been cited.[1] The patient is premenopausal at 41 years old, within the range of known causes, but younger than that in which the majority of these tumors occur.

### Clinical presentation

The majority (22%–44%) of struma ovarii cases are asymptomatic. However, the most common clinical symptom is pelvic pain, followed by the presence of a pelvic mass.[1] Other nonspecific symptoms such as abdominal pain, back pain, vaginal bleeding, and frequent urination have been described.[4] Five–eight percent (5%–8%) of masses have functional thyroid tissues causing hyperthyroidism in the absence of thyroid gland enlargement.[1] The index patient initially presented with a pelvic mass associated with abdominal enlargement due to massive ascites.

The etiology of ascites remains speculative. A hypothesis was that the physical contact of the solid ovarian tumor and peritoneum cause mechanical irritation and peritoneal stimulation, thereby stimulating peritoneal inflammation and overproduction of peritoneal fluid.[6] Another theory suggested that tumor compression on the underlying lymph vessels and veins results to the

slow reabsorption of normal peritoneal fluid, decrease in lymphatic and venous drainage, and escape of fluid through the superficial lymphatics of the tumor.[2] Hypoalbuminemia, as seen in this case, also contributed to the fluid accumulation of excessive fluid in the peritoneum due to a decrease in oncotic pressure.[3]

The index patient also presented with pleural effusion. The most established mechanism of pleural effusion is the mechanical transference of excessive ascitic fluid into the pleural cavity through tiny defects in the diaphragm.[3] The negative pressure within the pleural space with the high abdominal pressure accompanying ascites raises blebs on the superior surface of the diaphragm resulting to defects.[6] Chest and abdominal scans after intraperitoneal injection of contrast confirmed peritoneal-to-pleural fluid flow.[3] Biochemical studies showed that the composition of pleural and peritoneal fluids is identical. The right-sided pleural effusion is classically more common due to the larger diameter in the right lymphatic openings. However, the left-sided pleural effusion present in the patient and bilateral pleural effusions have been reported.[6]

Because of the primary consideration of ovarian malignancy, serum marker CA-125 was requested and was markedly elevated in the index patient. However, CA-125 elevation may also be associated with nonmalignant conditions such as benign masses, pregnancy, endometriosis, and tuberculosis.[2,7] This makes CA-125 relatively nonspecific specially for premenopausal women.[8] The exact reason for the elevated tumor marker in PMS remains unclear. A proposed explanation is the irritation and inflammation of pleural and peritoneum surfaces produced by the presence of free fluid in these spaces.[2]

Another unique feature was the concomitant abdominopelvic tuberculosis. This type of tuberculosis could also mimic advanced ovarian malignancy. The clinical triad of abdominal pain, ascites, and adnexal mass along with raised CA-125 levels should raise the suspicion of pelvic tuberculosis, especially in countries with high prevalence like the Philippines.[7] In this case, the signs and symptoms presented were consistent with the aforementioned clinical picture. A review of the literature (MEDLINE search terms: pseudo-Meigs' and tuberculosis) documented only one case of PMS with abdominal tuberculosis.[8] This case report in India narrated a postmenopausal woman who underwent surgical management before tuberculosis treatment resulting to full postoperative resolution. Despite anti-Koch's administration before surgery in the index case, there was reaccumulation of ascites suggesting that surgical intervention is necessary for full remission.

## Diagnosis

A definite approach to the diagnosis of struma ovarii-related PMS has not been established, and an accurate preoperative diagnosis still remains a medical dilemma.[4] Imaging procedures of ultrasound and CT scan remained to be helpful to determine the characteristic and extent of the mass, involvement of adjacent structures, and exclude metastatic disease before treatment. On transvaginal ultrasonography, struma ovarii typically appears as cystic tumors containing one or more well-circumscribed round solid areas with smooth contour known as "struma pearls." CT scan manifestations are cystic-solid mass, lobulated shape with clear boundaries.[4] These characteristics were manifested by the diagnostic imaging done for the case.

Surgery remains to be the definitive treatment. Incorrectly diagnosed cases, mostly as ovarian malignancy, lead to unnecessary extensive surgeries.[9] The patient underwent gynecologic surgery with staging. In the cases with a high index of suspicion, intraoperative diagnosis by frozen section, which was not performed in this case, could be done to confirm the benign nature of the mass.[2,3,5] The accuracy of intraoperative frozen section ranges from 80% to 90%.[3] The majority of the cases do not undergo thyroid function tests because the patients were asymptomatic for any thyroid symptoms.[4,10] The usefulness of thyroid scintigraphy or PET using iodine or 99 m Tc-pertechnetate has shown promising outcomes in identifying struma ovarii with PMS that mimics advanced ovarian cancer.[9]

Grossly, the tumor in struma ovarii is described as solid, lobulated, and predominantly cystic, sometimes confused as serous cystadenoma. The average size and laterality of the mass were noted to be 10 cm in the largest dimension and mostly unilateral with the right-sided predominance but with 9.1% of cases reported to be bilateral.[2] In the index patient, intraoperative findings showed a right ovarian multilobulated mass measuring 13.0 cm × 16.0 cm × 6.0 cm with solid-cystic structures measuring 9.5 cm in greatest tumor dimension [Figure 4], similar to the usual description of a struma ovarii. The liver, subdiaphragmatic surface, spleen, kidneys, stomach, intestines, gallbladder, and appendix were smooth and grossly normal. There were no palpable pelvic or paraaortic lymph nodes. The final histopathologic result was struma ovarii with the presence of multiple thyroid macrofollicles filled with colloid, negative for malignancy [Figure 5].

## Management and follow-up

In the cases of PMS presenting with dyspnea, the patients could undergo paracentesis or thoracentesis for symptomatic relief. Yet, the mainstay treatment is

surgical excision of ovarian mass. With the consideration of malignancy in the index patient, exploratory laparotomy, total abdominal hysterectomy, and bilateral salpingo-oophorectomy with paraaortic lymph node sampling were performed.

The controversy remains regarding whether radical or conservative surgery should be practiced and what the optimum approach of surgical access (laparotomy vs. laparoscopy) should be.[3] In women of reproductive age, fertility-conserving surgery such as unilateral salpingo-oophorectomy or cystectomy is the treatment of choice. On the other hand, in postmenopausal women, total abdominal hysterectomy with bilateral salpingo-oophorectomy is warranted. Studies showed that access through laparoscopy is now established, technically feasible for struma ovarii, and is comparable to laparotomy approach. Certain disadvantages in the removal of a potentially malignant ovarian mass through laparoscopy include decreased accuracy of staging, increased possibility of port-site metastasis, and increased risk of spillage of cancerous cells into the abdominal cavity. However, the laparoscopic approach shortened the length of hospital stay and did not negatively impact disease-free or overall survival rates of patients.[10]

Struma ovarii cases classified either as benign or malignant without metastases have an excellent prognosis.[4] Complete resolution of pleural effusion and ascites with normalization of CA-125 levels are observed after tumor removal without any adjuvant therapy.[1,3,5] The patient's postoperative course was unremarkable with a total resolution of the ascites and pleural effusion. With the additional diagnosis of abdominopelvic Koch's, the standard treatment with a 6-month anti-Koch's regimen was completed.

No consensus on the interval or length of follow-up has been standardized for struma ovarii. Based on the studies reported in the literature, for the malignant types of struma ovarii, initial recurrence was reported after an average of 4 years.[5] Long-term follow-up was required in these cases. On the other hand, for the majority of benign cases, as in the index patient, long-term follow-up was not considered. The index patient has been seen once at the general clinic and twice through teleconsultation for follow-up with no recurrence of symptoms.

## Summary

Increasing awareness of the occurrence of benign complex pelvic masses, such as struma ovarii, which mimic malignant conditions based on clinical, ultrasound, and/or biochemical features, is essential in reaching for a correct diagnosis. Physicians should be knowledgeable of the differential diagnostics needed to determine what type of mass they are dealing with. Thus, considering PMSand carrying out laboratory examinations such as cytology and frozen section could aid the clinicians in accurately diagnosing and planning surgical management appropriate for the patient.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

## Conflicts of interest

There are no conflicts of interest.

## References

1. Morales OA, Araiza JJ, Alvarez Dominguez TP, Guzman R, Barbabosa JA, Tolentino Sosa MI. Struma ovarii associated with pseudo-Meigs' syndrome and elevated serum ca 125: Case report and literature review. Int J Reprod Contracept Obstet Gynecol 2017;7:336.
2. Jin C, Dong R, Bu H, Yuan M, Zhang Y, Kong B. Coexistence of benign struma ovarii, pseudo-Meigs' syndrome and elevated serum CA 125: Case report and review of the literature. Oncol Lett 2015;9:1739-42.
3. Yadav S, Tomar R, Verma N, Khurana N, Triathi R. Struma ovarii with pseudo-Meigs' syndrome and raised cancer antigen-125 levels masquerading as an ovarian carcinoma case report and literature review. Sultan Qaboos Univ Med J 2017;17:e229-33.
4. Zhang T, Chen P, Gao Y. Struma ovarii: A mini review. Int J Clin Exp Med 2018;11:10364-71. Available from: http://www.ijcem.com/files/ijcem0070646.pdf. [Last accessed on 2021 Jul 13].
5. Reddy AS, Babu H, Tsama LS, Narmadha NS. Pseudo-Meigs syndrome with ovarian and abdominal tuberculosis: A case report. Biomed J Sci Tech Res 2017;1:(5). [Doi: 10.26717/bjstr.2017.01.000471].
6. Fujiwara S, Tsuyoshi H, Nishimura T, Takahashi N, Yoshida Y. Precise preoperative diagnosis of struma ovarii with pseudo-Meigs' syndrome mimicking ovarian cancer with the combination of $^{131}$I scintigraphy and $^{18}$F-FDG PET: Case report and review of the literature. J Ovarian Res 2018;11:11.
7. Miyoshi A, Miyatake T, Hara T, Tanaka A, Komura N, Komiya S, *et al.* Etiology of ascites and pleural effusion associated with ovarian tumors: Literature review and case reports of three ovarian tumors presenting with massive ascites, but without peritoneal dissemination. Case Rep Obstet Gynecol 2015;2015:414019.
8. Reddy AS, Babu H, Tsama LS, Narmadha NS. Pseudo-Meigs syndrome with ovarian and abdominal tuberculosis: A case report. Biomed J Sci Tech Res 2017;1:(5). [Doi: 10.26717/bjstr.2017.01.000471].
9. Imtiaz S, Siddiqui N, Ahmad M, Jahan A. Pelvic-peritoneal tuberculosis mimicking ovarian cancer. J Coll Physicians Surg Pak 2012;22:113-5.
10. Wee JY, Li X, Chern BS, Chua IS. Struma ovarii: Management and follow-up of a rare ovarian tumour. Singapore Med J 2015;56:35-9.