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Giant struma ovarii with pseudo-Meigs'syndrome and raised cancer antigen-125 levels: A case report

Yan Liu, Gao-Yan Tang, Lu Liu, Hui-Min Sun, Hai-Yan Zhu

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Abstract

BACKGROUND

Struma ovarii is a type of monodermal mature teratoma composed entirely or mainly of thyroid tissue, accounting for 1% to 3% of all ovarian teratomas and 0.3% to 1.0% of all ovarian tumors. Of which, struma ovarii with ascites and pleural effusion, called pseudo-Meigs'syndrome and raised cancer antigen-125 levels (CA 125) is even rarer.

CASE SUMMARY

This paper reports the diagnosis and treatment of a patient of struma ovarii with pseudo-Meigs'syndrome, presenting with the clinical features of ovarian carcinoma: Complex pelvic mass, gross ascites, right pleural effusion and markedly elevated serum CA 125 levels. During the operation, a cystic-solid mass about 20 cm × 10 cm × 5 cm in the right adnexa and a solid mass with the size of 3 cm × 2 cm × 0.1 cm in the left ovary were observed. She underwent right adnexectomy and resection of the left ovarian mass and histopathology revealed a mature left-sided ovarian teratoma and struma ovarii of right adnexal mass. During 1-year follow-up, the patient recovered well, tumor markers and other indicators returned to normal.

CONCLUSION

The diagnosis and treatment process of this case suggests that the clinical symptoms of struma ovarii with pseudo-Meigs'syndrome are lack specificity, which is easily misdiagnosed. Clinicians should improve the understanding of this disease, enhance the awareness of early screening, and improve the level of diagnosis and treatment.

Key Words: Struma ovarii; pseudo-Meigs'syndrome; Ascites; Pleural effusion; Cancer antigen-125; Case report

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Core Tip: Struma ovarii with pseudo-Meigs'syndrome and elevated serum cancer antigen-125 is easily preoperatively misdiagnosed as ovarian cancer, leading to unnecessary extended surgery. In this case, the patient of giant struma ovarii with pseudo-Meigs'syndrome underwent conservative surgery in the form of a right salpingo-oophorectomy, as there was no evidence of malignancy according to the preoperative biopsy and intraoperative frozen analysis. Besides, this patient was premenopausal and to our knowledge, she is the youngest with this disease.

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INTRODUCTION

Struma ovarii, a special type of ovarian teratoma, is a highly differentiated monodermal teratoma that arises from ovarian primordial germ cells and is defined as mature teratoma composed of a minimum of 50% of thyroid tissue by World Health Organisation, accounting for 1% to 3% of all ovarian teratomas and 0.3% to 1.0% of all ovarian tumors[1,2]. Meigs'syndrome represents a solid benign ovarian neoplasm, such as fibroma, granulosa cell tumor or thecoma with hydrothorax and ascites which are completely resolved spontaneously after surgical removal of the tumour[3]. When ascites and hydrothorax are associated with other ovarian tumors, it is defined as pseudo-Meigs'syndrome[4]. Struma ovarii is rare, but struma ovarii with pseudo-Meigs'syndrome is even rarer and it is easily misdiagnosed in clinical practice[5]. In order to deepen clinicians' understanding of this disease, here, we present a case of benign struma ovarii associated with pseudo-Meigs'syndrome and elevated cancer antigen-125 (CA 125).

CASE PRESENTATION

Chief complaints

A 37-year-old, Chinese woman, premenopausal, presented to gynecologic clinic with a complaint of abdominal bulge for 4 mo.

History of present illness

Symptoms started 4 mo before presentation with abdominal bulge, without abdominal pain.

History of past illness

She had a history of breast fibroma surgery 6 years ago.

Personal and family history

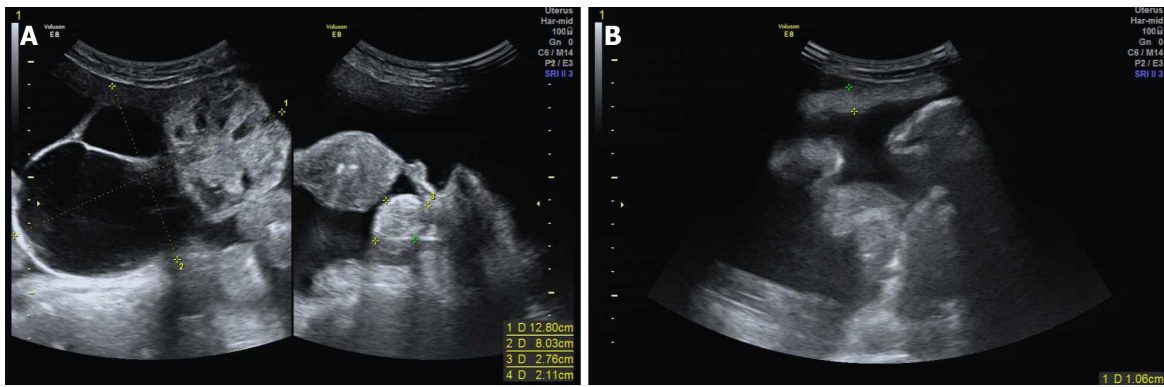
The patient denied any family history of malignant tumours.

Physical examination

Physical examination revealed obvious abdominal distension, positive mobility voiced sounds, positive fluid wave tremor and weak bowel sounds. Besides, the vital signs were as follows: Body temperature, 37.2 °C; blood pressure, 122/83 mmHg; pulse, 102 beats per min; respiratory rate, 18 breaths per min. Furthermore, the right breast had old surgical scars. Gynecological examination: an irregular mass, with a diameter of 12 cm, was found on the right ovary; left ovary and uterus had no obvious abnormalities.

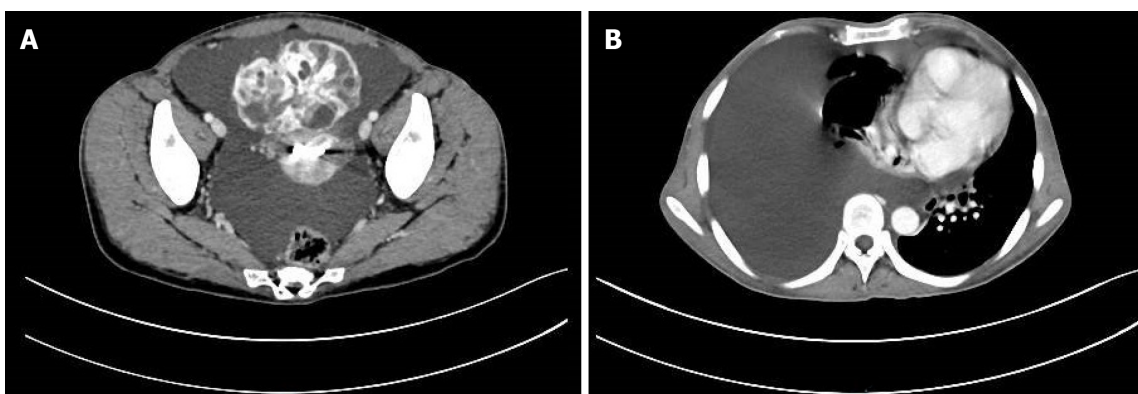
Laboratory examinations

Tumor marker carbohydrate antigen 199 was not elevated (33.87 U/mL, reference, 0-37), but CA 125 was 1492.23 U/mL (reference, 0-35). Besides, thyroid function tests were within normal limits: free triiodothyronine, 6.24 pmol/L (reference, 3.5-6.5); free thyroxine, 19.63 pmol/L (reference, 11.5-22.7);



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Figure 1 Transvaginal ultrasound images. A: Transvaginal ultrasound (US) demonstrating a large (12.8 cm × 8.0 cm), solid, cystic mass, originating in the right ovary and a small (2.8 cm × 2.1 cm), solid left adnexal mass; B: US showing a large amount of free peritoneal fluid and thickened greater omentum.



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Figure 2 Computed tomography scan imaging of the chest and pelvis. A: Computed tomography (CT) scan of the chest revealing right lung atelectasis with a large right pleural effusion; B: CT scan of the pelvis showing gross ascites and a large complex cystic pelvic mass.

thyroid-stimulating hormone, 1.44 $\mu\text{IU}/\text{mL}$, (reference, 0.55-4.78). No abnormality was found in routine blood analyses.

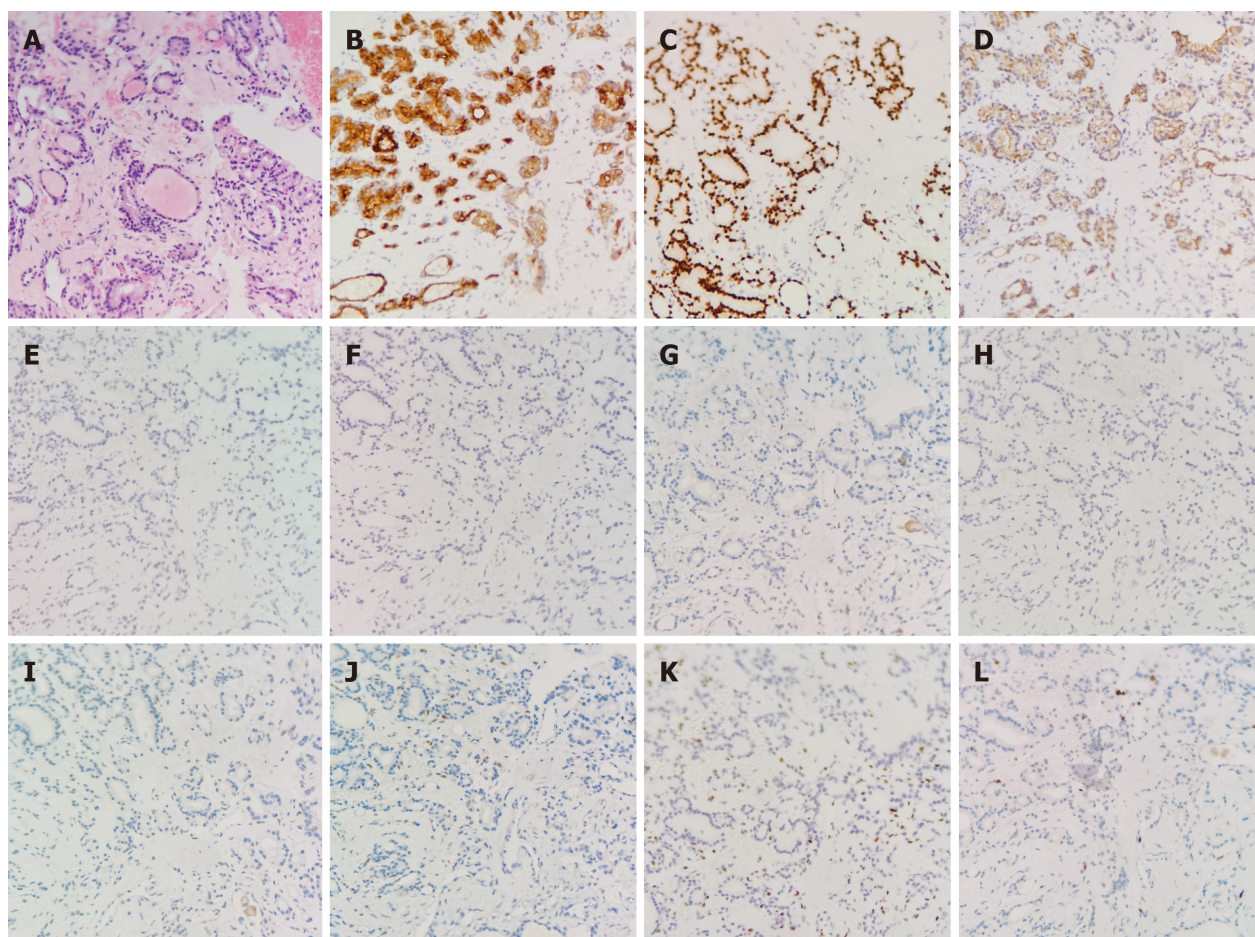
Imaging examinations

Ultrasonography showed a 12.8 cm × 8.0 cm right adnexal mass containing solid and cystic components with abundant vascularization and 2.8 cm × 2.1 cm solid left adnexal mass. Besides, there was a large amount of free peritoneal fluid and thickened greater omentum (Figure 1). Computed tomography (CT) scan of the chest, abdomen, and pelvis revealed right lung atelectasis with a large right pleural effusion, gross ascites, and a large complex cystic pelvic mass (Figure 2). Overall, the radiological findings were suspicious of ovarian cancer.

FINAL DIAGNOSIS

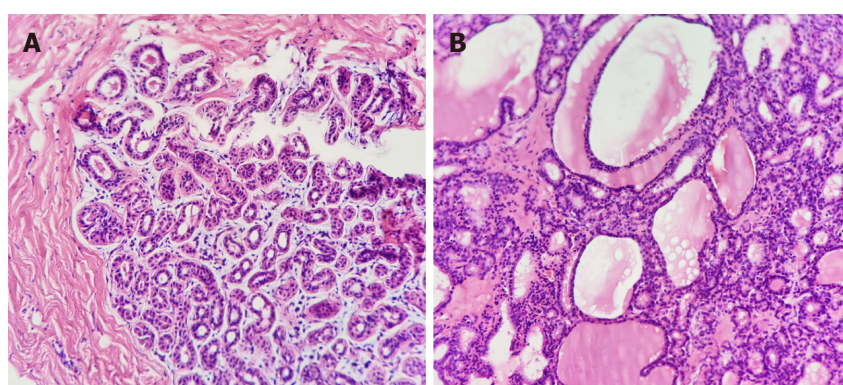
Cytological examination of pleural fluid and ascites indicated only reactive mesothelial cells with a few lymphocytes, histiocytes and neutrophils with no malignant cells identified. Then pathological histology of percutaneous biopsy of the pelvic mass showed hyperplastic fibrous tissue and mature thyroid follicles, without cellular and structural atypia, which was suspicious of struma ovarii combined with immunohistochemistry (Figure 3). Combined with the analysis of pathological histology and immunohistochemistry of the biopsies, the preoperative diagnosis was highly suspicious of struma ovarii.

The final histopathology revealed a mature left-sided ovarian teratoma and struma ovarii of right adnexal mass (Figure 4).



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Figure 3 Hematoxylin-eosin staining and immunohistochemistry of percutaneous biopsy of the pelvic mass. A: Hematoxylin-eosin staining showed multiple benign colloid-filled thyroid follicles ($\times 200$); B-D: Immunohistochemistry (IHC) examination revealed that pelvic mass was positive for thyrolobulin ($\times 200$), thyroid transcription factor-1 ($\times 200$) and Cytokeratin-7 ($\times 200$), respectively; E-L: IHC examination revealed that pelvic mass was negative for Cytokeratin-20 ($\times 200$), caudal-related homeobox transcription factor 2 ($\times 200$), Estrogen receptor ($\times 200$), calretinin ($\times 200$), P53 ($\times 200$), P16 ($\times 200$), Wilms tumor-1 ($\times 200$) and Ki-67 ($\times 200$), respectively.



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Figure 4 Histopathological analysis of the resected specimen. A: Hematoxylin-eosin stained left ovary showing teratoma ($\times 200$); B: Microscopic appearance of the right ovary showing variable-sized thyroid follicles ($\times 200$).

TREATMENT

The patient was arranged for an exploratory laparotomy for diagnostic and therapeutic purposes on October 22, 2020. During the operation, 3000 mL of straw-colored ascites was drained. A large solid neoplasm (20 cm \times 10 cm \times 5 cm) originating from the right ovary was twisted clockwise for half a turn together with the right fallopian tube and part of the intestinal canal was adherent to the mass. Besides,

the left ovary was slightly atrophic, containing a cystic mass, with the size of 3 cm × 2 cm × 0.1 cm. Intraoperative examination of all abdominal and pelvic organs did not show any additional lesions. The patient subsequently underwent right salpingo-oophorectomy and resection of the left ovarian mass and intestinal adhesiolysis and the excised specimens were sent for frozen analysis to rule out malignancy.

OUTCOME AND FOLLOW-UP

The patient recovered uneventfully and pleural effusion disappeared 5 d after surgery. Besides, CA 125 returned to normal range level (27.26 U/mL) 1 mo after surgery. The patient was followed up for 1 year after operation and there were no signs of obvious abnormality.

DISCUSSION

Struma ovarii, as a highly specific mature teratoma, is mostly benign, with malignant transformation only occurring in 0.5%-10 % of cases[1,6]. Struma ovarii can occur in female patients of any age, but perimenopause is the peak period of the disease, and it is usually asymptomatic, whereas patients with large struma ovarii may show abdominal distention, as in our case[7]. Because there are no obvious specificities in ultrasound, CT or magnetic resonance imaging (MRI) for struma ovarii, it is difficult to differentiate from ovarian cancer on imaging, especially for struma ovarii accompanied by ascites and pleural effusion, called pseudo-Meigs'syndrome and elevated CA 125, which can mimic ovarian malignancy. Fujiwara *et al*[8] reported positron emission tomography/CT combined with thyroid scintigraphy may be useful to define the diagnosis in struma ovarii with pseudo-Meigs'syndrome. Up to now, accurate preoperative diagnosis for struma ovarii by conventional imaging alone remains challenging and postoperative pathology is still required to confirm the diagnosis.

In the literature, 13 cases have been published on struma ovarii combined with pseudo-Meigs'syndrome and elevated CA 125, we describe another case of struma ovarii combined with pseudo-Meigs'syndrome and elevated CA 125[5,8-18]. Of all the cases, most of the patients were in their fifth or sixth decade when diagnosed with struma ovarii and almost 78.6% (11/14) of cases were postmenopausal women. Then, the most common presenting symptom was abdominal distention and the tumour sizes ranged between 5-23 cm in the large dimension, with an average size of 12 cm. Besides, most cases were preoperatively misdiagnosed as ovarian cancer and were treated by hysterectomy and bilateral salpingo-oophorectomy[5,8-17] and only two patients (including this case) underwent conservative surgery[18]. The ascites and hydrothorax disappeared completely and CA 125 levels returned to normal following surgery and all the cases had good prognosis. There are some unique features in our patient. Firstly, she was premenopausal, and to our knowledge, she is the youngest with this disease. Secondly, considering young age of our patient in order to avoid postoperative hormonal substitution, she underwent conservative surgery in the form of a right salpingo-oophorectomy as there was no evidence of malignancy according to analysis of percutaneous biopsy of the pelvic mass and frozen examination.

CONCLUSION

In summary, we confirm that struma ovarii is difficult to characterize on conventional imaging modalities and such patients should be diagnosed based on imaging features combined with pathology. In addition, more precise preoperative diagnosis should be performed to avoid unnecessary extended surgery.

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FOOTNOTES

Author contributions: Tang GY wrote the first draft of the manuscript; Liu Y and Liu L were responsible for treatment of the patient; Sun HM provided images of hematoxylin-eosin and immunohistochemistry; Zhu HY revised the manuscript; all authors have read and approved the final manuscript.

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