Contents

REVIEW

10823 New insights into the interplay between intestinal flora and bile acids in inflammatory bowel disease
Zheng L

10840 Role of visfatin in obesity-induced insulin resistance
Abdalla MMI

MINIREVIEWS

10852 Hyperthermic intraperitoneal chemotherapy and colorectal cancer: From physiology to surgery

10862 New-onset diabetes secondary to acute pancreatitis: An update
Yu XQ, Zhu Q

10867 Ketosis-prone diabetes mellitus: A phenotype that hospitalists need to understand
Boike S, Mir M, Rauf I, Jama AB, Sunesara S, Mushtaq H, Khedr A, Nitesh J, Surani S, Khan SA

10873 2022 Monkeypox outbreak: Why is it a public health emergency of international concern? What can we do to control it?
Ren SY, Li J, Guo RD

ORIGINAL ARTICLE

Retrospective Cohort Study

10882 Clinical characteristics and prognosis of non-small cell lung cancer patients with liver metastasis: A population-based study

Retrospective Study

10896 Prevalence and risk factors for Candida esophagitis among human immunodeficiency virus-negative individuals
Chen YH, Jao TM, Shiue YL, Feng JJ, Hsu PI

10906 Prognostic impact of number of examined lymph nodes on survival of patients with appendiceal neuroendocrine tumors
Du R, Xiao JW

Observational Study

10921 Clinical and epidemiological features of ulcerative colitis patients in Sardinia, Italy: Results from a multicenter study
<table>
<thead>
<tr>
<th>Page</th>
<th>Title</th>
<th>Authors</th>
</tr>
</thead>
<tbody>
<tr>
<td>10931</td>
<td>Clinical observation of laparoscopic cholecystectomy combined with endoscopic retrograde cholangiopancreatography or common bile duct lithotripsy</td>
<td>Niu H, Liu F, Tian YB</td>
</tr>
<tr>
<td>10939</td>
<td>Patient reported outcome measures in anterior cruciate ligament rupture and reconstruction: The significance of outcome score prediction</td>
<td>Al-Dadah O, Shepstone L, Donell ST</td>
</tr>
<tr>
<td>10956</td>
<td>Body mass index and outcomes of patients with cardiogenic shock: A systematic review and meta-analysis</td>
<td>Tao WX, Qian GY, Li HD, Su F, Wang Z</td>
</tr>
<tr>
<td>10967</td>
<td>Impact of being underweight on peri-operative and post-operative outcomes of total knee or hip arthroplasty: A meta-analysis</td>
<td>Ma YP, Shen Q</td>
</tr>
<tr>
<td>10984</td>
<td>Branched-chain amino acids supplementation has beneficial effects on the progression of liver cirrhosis: A meta-analysis</td>
<td>Du JY, Shu L, Zhou YT, Zhang L</td>
</tr>
<tr>
<td>10997</td>
<td>Wells’ syndrome possibly caused by hematologic malignancy, influenza vaccination or ibrutinib: A case report</td>
<td>Šajn M, Luzar B, Zver S</td>
</tr>
<tr>
<td>11004</td>
<td>Giant cutaneous squamous cell carcinoma of the popliteal fossa skin: A case report</td>
<td>Wang K, Li Z, Chao SW, Wu XW</td>
</tr>
<tr>
<td>11010</td>
<td>Right time to detect urine iodine during papillary thyroid carcinoma diagnosis and treatment: A case report</td>
<td>Zhang SC, Yan CJ, Li YF, Cui T, Shen MP, Zhang JX</td>
</tr>
<tr>
<td>11031</td>
<td>Neonatal Cri du chat syndrome with atypical facial appearance: A case report</td>
<td>Bai MM, Li W, Meng L, Sang YF, Cui YJ, Feng HY, Zong ZT, Zhang HB</td>
</tr>
<tr>
<td>11037</td>
<td>Complete colonic duplication presenting as hip fistula in an adult with pelvic malformation: A case report</td>
<td>Cai X, Bi JT, Zheng ZX, Liu YQ</td>
</tr>
<tr>
<td>Page</td>
<td>Title</td>
<td>Authors</td>
</tr>
<tr>
<td>------</td>
<td>------------------------------------------------------------------------------------------------</td>
<td>---------------------------------------------------------------------------------------------</td>
</tr>
<tr>
<td>11044</td>
<td>Autoimmune encephalitis with posterior reversible encephalopathy syndrome: A case report</td>
<td>Dai SJ, Yu QJ, Zhu XY, Shang QZ, Qu JB, Ai QL</td>
</tr>
<tr>
<td>11059</td>
<td>Different intraoperative decisions for undiagnosed paraganglioma: Two case reports</td>
<td>Kang D, Kim BE, Hong M, Kim J, Jeong S, Lee S</td>
</tr>
<tr>
<td>11074</td>
<td>Bone marrow metastatic neuroendocrine carcinoma with unknown primary site: A case report and review of the literature</td>
<td>Shi XB, Dong WX, Jin FX</td>
</tr>
<tr>
<td>11101</td>
<td>Severe Klebsiella pneumoniae pneumonia complicated by acute intra-abdominal multiple arterial thrombosis and bacterial embolism: A case report</td>
<td>Bao XL, Tang N, Wang YZ</td>
</tr>
<tr>
<td>11111</td>
<td>Spontaneous bilateral femur neck fracture secondary to grand mal seizure: A case report</td>
<td>Senocak E</td>
</tr>
<tr>
<td>11116</td>
<td>Favorable response after radiation therapy for intraductal papillary mucinous neoplasms manifesting as acute recurrent pancreatitis: A case report</td>
<td>Harigai A, Kume K, Takahashi N, Omata S, Umezawa R, Jingu K, Masamune A</td>
</tr>
<tr>
<td>11139</td>
<td>Perirectal epidermoid cyst in a patient with sacrococcygeal scoliosis and anal sinus: A case report</td>
<td>Ji ZX, Yan S, Gao XC, Lin LF, Li Q, Yao Q, Wang D</td>
</tr>
<tr>
<td>Page</td>
<td>Title</td>
<td>Authors</td>
</tr>
<tr>
<td>------</td>
<td>----------------------------------------------------------------------</td>
<td>----------------------------------------------</td>
</tr>
<tr>
<td>11162</td>
<td>Longest survival with primary intracranial malignant melanoma: A case report and literature review</td>
<td>Wong TF, Chen YS, Zhang XH, Hu WM, Zhang XS, Lv YC, Huang DC, Deng ML, Chen ZP</td>
</tr>
<tr>
<td>11172</td>
<td>Spontaneous remission of hepatic myelopathy in a patient with alcoholic cirrhosis: A case report</td>
<td>Chang CY, Liu C, Duan FF, Zhai H, Song SS, Yang S</td>
</tr>
<tr>
<td>11178</td>
<td>Cauda equina syndrome caused by the application of DuraSealTM in a microlaminectomy surgery: A case report</td>
<td>Yeh KL, Wu SH, Fuh CS, Huang YH, Chen CS, Wu SS</td>
</tr>
<tr>
<td>11185</td>
<td>Bioceramics utilization for the repair of internal resorption of the root: A case report</td>
<td>Riyahi AM</td>
</tr>
<tr>
<td>11198</td>
<td>Accidental esophageal intubation via a large type C congenital tracheoesophageal fistula: A case report</td>
<td>Hwang SM, Kim MJ, Kim S, Kim S</td>
</tr>
<tr>
<td>11204</td>
<td>Ventral hernia after high-intensity focused ultrasound ablation for uterine fibroids treatment: A case report</td>
<td>Park JW, Choi HY</td>
</tr>
</tbody>
</table>

**LETTER TO THE EDITOR**

<table>
<thead>
<tr>
<th>Page</th>
<th>Title</th>
<th>Author</th>
</tr>
</thead>
<tbody>
<tr>
<td>11210</td>
<td>C-Reactive protein role in assessing COVID-19 deceased geriatrics and survivors of severe and critical illness</td>
<td>Nori W</td>
</tr>
</tbody>
</table>
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Giant struma ovarii with pseudo-Meigs’s syndrome and raised cancer antigen-125 levels: A case report

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

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’s 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’s 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’s 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’s syndrome; Ascites; Pleural effusion; Cancer antigen-125; Case report

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Core Tip: Struma ovarii with pseudo-Meigs’s 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’s 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.

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’s 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’s syndrome[4]. Struma ovarii is rare, but struma ovarii with pseudo-Meigs’s 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’s 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);
thyroid-stimulating hormone, 1.44 μIU/mL, (reference, 0.55-4.78). No abnormality was found in routine blood analyses.

**Imaging examinations**

Ultrasoundography showed a 12.8 cm × 8.0 cm right adnexal mass containing solid and cystic components with abundant vascularity 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).
Figures 3 and 4 illustrate the histopathological analysis of the pelvic mass and the resected specimen, respectively. The pelvic mass was found to be composed of multiple benign colloid-filled thyroid follicles, as evidenced by hematoxylin-eosin staining. Immunohistochemistry examination revealed positivity for thyroblastin, thyroid transcription factor-1, and cytokeratin-7, among other markers. Conversely, the pelvic mass was negative for cytokeratin-20, caudal-related homeobox transcription factor 2, estrogen receptor, calretinin, p53, p16, Wilms tumor-1, and Ki-67.

The resected specimen showed a teratoma of the left ovary and variable-sized thyroid follicles in the right ovary, as observed under hematoxylin-eosin staining.

**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 × 10 cm × 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’s 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’s 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’s syndrome and elevated CA 125, we describe another case of struma ovarii combined with pseudo-Meigs’s 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 distension 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: TangGY wrote the first draft of the manuscript; LiuY and LiuL 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.
Liu Y. Struma ovarii with pseudo-Meigs’ syndrome

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